Genital perception and health-related quality of life of adults operated for hypospadias

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by

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Abstract

Hypospadias is a common penile malformation, which is usually surgically corrected in early childhood. Severe types of hypospadias are corrected to promote a normal sexuality and micturition, whereas mild types are mainly operated to improve appearance. There are only a few studies, which have examined the long-term outcomes of hypospadias repair. The aims of the present PhD project were, first (study 1), to assess how men and women perceive surgically corrected genitals of men with hypospadias in comparison with circumcised genitals. In addition, the most relevant predictors of the perception of these genitals were evaluated. Moreover, we studied how women consider single aspects of penile appearance. Second (study 2), we sought to evaluate the health-related quality of life (HRQoL) of men with a corrected hypospadias in comparison to circumcised men and to examine determinants of hypospadias patients' HRQoL.

In summary, the present research project indicates that women and men perceived the penile appearance of men with operated mild types of hypospadias as satisfactory and as normal-looking as circumcised genitals. In addition, women were found to consider the position and shape of the urethral opening as the least important aspect of penile appearance. Since urethral reconstruction to reposition the urethral opening is one of the key issues in mild hypospadias, these findings may stimulate reflections regarding the relevance of surgical correction of mild types of hypospadias in early childhood. Finally, our study found a normal HRQoL of men with corrected hypospadias. However, a negative genital self-perception was found to be a risk factor for an impaired mental HRQoL.

Abbreviations

CB model: Cognitive-Behavioral model

EF: Erectile function

GPS: Genital Perception Score

HASRGs: Hypospadias-affected surgically-repaired genitals

HRQoL: Health-related quality of life

IIEF: International Index of Erectile Function

MCS: Mental component summary score of SF-36

OF: Orgasmic function

PPS: Penile Perception Score

PCS: Physical component summary score of SF-36

QoL: Quality of life

SES: Socio-economic status

SF-36: Medical Outcomes Study Short Form-36 item questionnaire

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Part A General Introduction

The aim of this doctoral thesis was to learn more about the long-term psychological outcome of hypospadias repair. First, we examined how surgically corrected genitals of men with hypospadias (a common congenital penile malformation) are evaluated by men and women without previous knowledge of hypospadias. Second, we extensively investigated health-related quality of life (HRQoL) in men with corrected hypospadias. This introductory chapter gives background information about the topic. It includes information about the medical basis of hypospadias (section A1), a review of the literature concerning genital perception (section A2) and HRQoL (section A3) of men with corrected hypospadias, a summary of the gaps and limitations of the current literature (section A4), and finally the most important background information about the present research project (section A5). Section B comprises three articles which summarize the most important findings of our research project. Finally, section C delivers a general discussion of the main findings and a conclusion regarding clinical implications and directions for future research.

1. Hypospadias

1.1. Definition & Classification

Hypospadias is one of the most common penile malformations (Manzoni, Bracka, Palminteri, & Marrocco, 2004). It is manifested as a misplacement of the urethral meatus, presumably due to arrested penile development. Depending on the stage at which the interruption in normal penile development occurs, the meatus may be located at the ventral surface of the penis or in the perineum (Figure 1). In most types of hypospadias, the prepuce is split ventrally. More severe forms of hypospadias are associated in particular with a curvature and dysplasia of the penis (Baskin & Ebbers, 2006; Warren & Snodgrass, 2011).

Most current classifications are based on the anatomical position of the external meatus relative to the penis or to nonpenile structures (e.g. scrotum or perineum). A more proximal meatus corresponds to a more severe type of hypospadias (Macedo, Rondon, & Ortiz, 2012). Thus, the severity of hypospadias can be classified as (Figure 1) distal (glandular, coronal, or subcoronal) in approximately 60-65% of hypospadias cases; middle (midpenile) in approximately 20-30% of hypospadias cases; and posterior or proximal (posterior penile, penoscrotal, scrotal, or perineal) in approximately 10-15% of hypospadias cases (De Win, Cuckow, Hoebeke, & Wood, 2012). However, a conclusive assessment of the severity of hypospadias and the curvature of the penis can only be assessed during surgical exploration. In addition, this classification is less appropriate in penoscrotal, scrotal and perineal types of hypospadias because the scrotum may be located as far as the distal penis in these cases (Macedo et al., 2012).

Most hypospadias occur as isolated malformations (Manzoni et al., 2004) and fertility is not affected in distal forms of an isolated hypospadias (Asklund et al., 2010). The most common association (approximately 7%) is an undescended testis (Warren & Snodgrass, 2011). Patients with hypospadias and undescended testis may have impaired fertility (Asklund et al., 2010). If other elements of a disturbed sexual development can be found, the patients are diagnosed as having a disorder of sexual development (DSD). In cases in which other malformations occur besides hypospadias, the presence of a syndromic condition is more likely (Warren & Snodgrass, 2011).

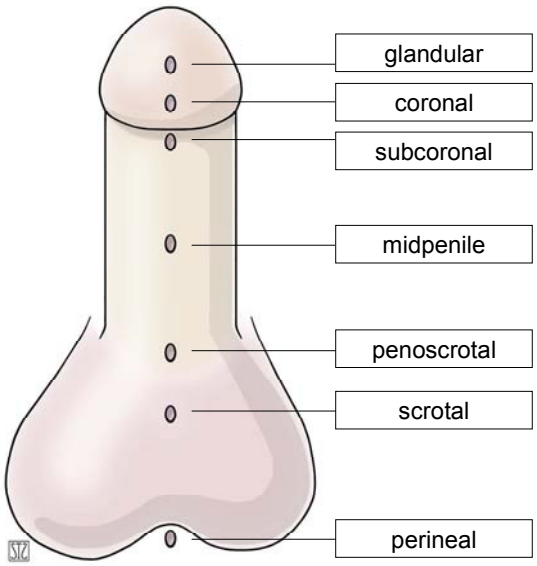

Figure 1: Hypospadias classification

(Susanne Staubli, University Children's Hospital Zurich)

1.2. Incidence & Etiology

Hypospadias occurs in approximately one of 200 to one of 300 live male births (Baskin & Ebbers, 2006; Manzoni et al., 2004). The possibility of an increasing prevalence of hypospadias in the last decades has been discussed. Some studies have found a growing incidence, for example, in Europe, whereas others could not confirm this observation in the same countries. However, as study data were collected differently, the results are difficult to compare. Some studies referred to hospital discharge registries, which only include surgically treated patients; others were based on birth defect surveillance systems. Furthermore, hypospadias may have been diagnosed and defined differently over time (van der Zanden et al., 2012).

Etiology is unknown in most hypospadias cases. As hypospadias shows familial clustering, genetic factors may play a role in familial hypospadias. Several genetic defects have been found (van der Zanden et al., 2012). As the development of the penis depends on the balance between androgens and estrogens some environmental factors have also been discussed (e.g. maternal exposure to synthetic estrogens). However, the current view is that the malformation

is most likely caused by environmental factors in genetically susceptible individuals by a mix of monogenetic and multifactorial forms involving both genes and environmental factors (van der Zanden et al., 2012). Although many studies have examined the role of environmental factors for the development of hypospadias, few factors have been found to be consistently involved in hypospadias: a low birth weight or being small for gestational age, placental insufficiency, maternal hypertension, pre-eclampsia, and exposure to maternal intrauterine diethylstilbestrol (DES), which is a synthetic form of estrogen (van der Zanden et al., 2012).

1.3. Surgical Treatment

A surgical correction of hypospadias has the following aims: first, a repositioning of the meatus to the tip of the glans to enable micturition in a standing position in a straight stream; second, correcting curvature to allow a normal sexual function and facilitate penetration; third, achieving a normal appearance of the penis (Aigrain, Cheikhelard, Lottmann, & Lortat-Jacob, 2010; Baskin & Ebbers, 2006; De Win et al., 2012; Manzoni et al., 2004).

Today, the trend in developed countries is to correct almost all types of hypospadias around the first birthday (Manzoni et al., 2004). Whereas severe proximal types of hypospadias are corrected to solve functional problems (e.g. curvature of the penis during erection), mild types of hypospadias are usually operated to achieve a better cosmetic result , as functional deficits are rare in mild forms (Fichtner, Filipas, Mottrie, Voges, & Hohenfellner, 1995; Manzoni et al., 2004). Today, hundreds of reconstructive techniques are available (Baskin & Ebbers, 2006; De Win et al., 2012). Only experienced specialists, pediatric urologists and pediatric surgeons, who often perform these operations, are qualified for hypospadias repair (Manzoni et al., 2004). Furthermore, a surgeon must be familiar with several methods for the correction of the different types of hypospadias (De Win et al., 2012). Complications that require secondary surgery may occur after hypospadias repair. Fistulas, stenosis and residual curvature of the penis are the most common (Aigrain et al., 2010; Baskin & Ebbers, 2006; De Win et al., 2012), accounting for a complication rate of 6% to 48% (De Win et al., 2012), depending on the severity of the hypospadias, the type of surgical technique , the surgeon's experience, and the length of follow-up (Aigrain et al., 2010; De Win et al., 2012). Nevertheless, hypospadias repair can be considered as safe, reliable and highly successful (Manzoni et al., 2004).

As mentioned above, most types of hypospadias are corrected at a very early age (around the first birthday) when a child is not yet able to decide. Violating physical integrity is accepted, because it is believed that early surgery optimizes patients' psychosexual development and quality of life (American Academy of Pediatrics, 1996). However, the positive effect of early

surgery on their psychosexual development is not evidence based. Therefore, studies on the long-term psychological outcome of hypospadias repair for instance studies regarding the health-related quality of life (HRQoL), are of crucial importance.

2. Genital perception of men with corrected hypospadias

2.1. The construct of genital perception

In my doctoral thesis, the term "genital perception" is used as a generic term for both genital self-perception, how satisfied a man is with various aspects of his penis and of his testicles, and genital perception by others, how satisfied a person is with various aspects of another man's penis or testicles. The terms "genital perception", "genital self-perception" and "genital perception by others" are based on the construct of "genital image" by Winter (1989), which is the construct regarding genital perception most widely used in the literature on men with no genital malformation. Winter's "genital image" construct is based on the concept of "body cathexis" by Secord and Jourard (1953, p. 343). The original term means "the degree of feeling of satisfaction or dissatisfaction with the various parts and processes of the body". The derived definition by Winter (1989, p. 4) describes the term "genital image" as "satisfaction with various aspects of the penis and testicles".

2.2. Application of the "genital perception" construct in the literature on genital perception of men with corrected hypospadias

A review of the literature on genital perception of men with corrected hypospadias shows that research does not refer to any construct or psychological theory of genital perception. However, most studies use self-developed, unvalidated questions to investigate genital self-perception that are nevertheless consistent with Winter's definition of genital image (Winter, 1989). To be more precisely, they examine, how satisfied each participant is with the appearance either of his penis or of several aspects of the penis.

In addition to self-developed questions, a few studies used sum scores to assess the genital perception of men with hypospadias-affected surgically-repaired genitals (HASRGs): For example, the "Genital Perception Score (GPS)" (Mureau, Slijper, Slob, & Verhulst, 1995) or the genital appraisal score (Mureau, Slijper, van der Meulen, Verhulst, & Slob, 1995) and the "Penile Perception Score (PPS)" (Weber, Landolt, Gobet, Kalisch, & Greeff, 2013). All scores evaluate patients' satisfaction with various aspects of the penis. In addition, the genital appraisal score also assesses whether subjects consider their penile appearance to be the same as in other men. The Penile Perception Score (PPS), the Genital Perception Score (GPS) and the genital appraisal score were calculated by adding the scores of each item.

The PPS by Weber et al. (2013) is not only an instrument to evaluate patients' genital self-perception. It is also a validated instrument for the outcome assessment of hypospadias repair by others (e.g. surgeons). In addition to the PPS, some other instruments objectively evaluate the outcome of hypospadias repair. For example, the Hypospadias Objective Scoring

Evaluation (HOSE) (Holland, Smith, Ross, & Cass, 2001) and the HOPE (Hypospadias Objective Penile Evaluation) – score (van der Toorn et al., 2013). The following chapters will first address genital self-perception of men with corrected hypospadias (chapter A 2.3.) and then genital perception of men with corrected hypospadias by others (chapter A 2.4.).

2.3. Genital self-perception of men with corrected hypospadias

2.3.1. Research on genital self-perception of men with corrected hypospadias

There are only a few studies that examine genital self-perception as long-term outcome of hypospadias repair. Furthermore, those investigations exhibit methodological limitations, such as a low response rate, heterogeneous patients, the use of self-developed, non-validated questionnaires and the lack of a control group (Rynja, de Jong, Bosch, & de Kort, 2011). Therefore, Rynja, de Jong, et al. (2011) conclude that any generalizations from the results and any conclusions must be made carefully.

Table 1 gives an overview of long-term studies that investigated the genital self-perception of men with corrected hypospadias. Studies were gathered with regard to the main questions of this doctoral thesis. Therefore, the investigations mentioned examine genital self-perception, satisfaction with penile appearance, concern of abnormal penile appearance or perceived difference in penile appearance from others, and satisfaction with penile size.

Studies which are not included in the table are, first, those which examine only the satisfaction with the surgical result (e.g. Heiss & Helmig, 1974; Leuthardt & Morger, 1987) and, second, studies which do not investigate long-term outcome but men's evaluation of their penile appearance shortly after hypospadias repair (e.g. after reoperation due to failed hypospadias repair in childhood) (e.g. Choi et al., 2013). A third group to be excluded contained studies involving participants who had hypospadias with sexual ambiguity (e.g. Callens et al., 2015; Eberle et al., 1993; Miller & Grant, 1997). A fourth group was studies that only investigated genital self-perception of children or adolescents (i.e. individuals younger than 18 years) (e.g. Jones, O'Brien, Chase, Southwell, & Hutson, 2009; Mureau, Slijper, Nijman, et al., 1995; Schonbucher, Landolt, Gobet, & Weber, 2008b; Weber, Schonbucher, Landolt, & Gobet, 2008) or participants who had a mean age lower than 16 years (e.g. Gray & Boston, 2003; Lam, Greenfield, & Williot, 2005; Nelson, Bloom, Kinast, Wei, & Park, 2005a; Seibold et al., 2010; Vandendriessche, Baeyens, Van Hoecke, Indekeu, & Hoebeke, 2010). Moreover, studies which do not contain any information (e.g. Hinderer, 2000; Svensson & Berg, 1983) or just unclear information regarding patients' mean age (e.g. Kvesic et al., 2005; Nuininga, DE Gier, Verschuren, & Feitz, 2005). Fifth, studies written in

languages other than German or English (e.g. Santamaría Ossorio & Sanjuán Rodríguez, 2002) were also excluded.

In line with the systematic review by Rynja, de Jong, et al. (2011), the present, more extensive study summary (Table 1) indicates that men with corrected hypospadias have a significantly worse genital self-perception (Mureau, Slijper, van der Meulen, et al., 1995; Mureau, Slijper, Slob, et al., 1995) and are significantly more dissatisfied with their genital appearance than control subjects (Aho, Tammela, Somppi, & Tammela, 2000; Kiss et al., 2011; Mondaini, Ponchietti, Bonafè, et al., 2002; Mureau, Slijper, Slob, et al., 1995; Wang et al., 2010; Wang et al., 2009). However, Moriya et al. (2006) and Mureau, Slijper, van der Meulen, et al. (1995) only found a tendency toward greater dissatisfaction with their genital appearance in men with corrected hypospadias. Furthermore, some men with HASRGs are concerned about having an abnormal penile appearance (Rynja, de Jong, et al., 2011), and significantly (Mureau, Slijper, van der Meulen, et al., 1995) or at least some more men with corrected hypospadias (Moriya et al., 2006) perceive their penile appearance to be different from that of other men than men without hypospadias. However, concerns about penile appearance are also mentioned by young men without genital malformations, and Rynja, de Jong, et al. (2011) conclude that men in general question their penile appearance.

Regarding penile size, men with corrected hypospadias reported being significantly (Mureau, Slijper, van der Meulen, et al., 1995; Rynja et al., 2009) or at least somewhat (Aho et al., 2000) more dissatisfied with their penile size than controls without hypospadias. Interestingly, satisfaction with penile length was found to correlate with satisfaction with penile appearance but not with objective, measured penile length (Rynja et al., 2009).

More results concerning genital self-perception in relation to several variables (such as medical factors) are listed in chapter 2.3.2.2.

Table 1: Overview of studies on genital self-perception of men with corrected hypospadias

Study references (authors)	**Participants (hypospadias group)**	**Controls**	**Measurement of four outcome-measures concerning genital self-perception**	**Outcome: Negative genital perception (measured with a sum score)**	**Outcome: Dissatisfaction with general penile appearance**	**Outcome: Concern of abnormal appearance/ Perceived difference in appearance from others**	**Outcome: Dissatisfaction with penile length**
Aho, Tammela, and Tammela (1997)	**N:** 43 **Mean-age:** 29.5 years **Age-range:** 20-38.6 years **Severity:** anterior, middle, posterior		self-developed questions		-		
Aho et al. (2000)	**N:** 48 **Mean-age:** 29.5 +/- 4.1 years **Severity:** anterior, middle, posterior	**N:** 43 age-matched, circumcised men **Mean-age:** 29.9 +/- 4.8 years	self-developed questions		**+c**		**=c**
Aulagne, Harper, de Napoli-Cocci, Bondonny, and Dobremez (2010)	**N:** 27 **Mean-age:** 25 years **Age-range:** 20-32 years **Severity:** proximal, scrotal		self-developed questions			+	**-**

Table 1 continued

Study references (authors)	**Participants (hypospadias group)**	**Controls**	**Measurement of four outcome-measures concerning genital self-perception**	**Outcome: Negative genital perception (measured with a sum score)**	**Outcome: Dissatisfaction with general penile appearance**	**Outcome: Concern of abnormal appearance/ Perceived difference in appearance from others**	**Outcome: Dissatisfaction with penile length**
Berg, Svensson, and Astrom (1981)	**N:** 34 **Mean-age:** 27.2 years **Age-range:** 21-34 years **Severity:** glandular, distal, mid and proximal penile, penoscrotal, perineal	**N:** 36 men operated on for verified appendicitis **Mean-age:** 26.9 years **Age-range:** 20-34 years	Interview with self-developed questions				**-** (no comparison with controls)
Bracka (1989)	**N:** 196 **Mean-age:** 22 years **Age-range:** 15-24 years **Severity:** distal, midshaft, proximal, short urethra		self-developed questions			**-**	

Table 1 continued

Study references (authors)	Participants (hypospadias group)	Controls	Measurement of four outcome-measures concerning genital self-perception	Outcome: Negative genital perception (measured with a sum score)	Outcome: Dissatisfaction with general penile appearance	Outcome: Concern of abnormal appearance/ Perceived difference in appearance from others	Outcome: Dissatisfaction with penile length
Chertin et al. (2013)	**N:** 119 **Age:** ≥ 18 years **Severity:** glanular, distal, proximal		self-developed questions		Glanular & distal: - Proximal: +		
Fraumann et al. (2014)	**N:** 13 **Mean-age:** 21 years **Age-range:** 18-30 years **Severity:** midshaft, proximal penile, penoscrotal, scrotal		self-developed questions		-		
Glassman, Machlus, and Kelalis (1980)	**N:** 101 **Mean-age:** 27 years **Age-range:** 16-68 years **Severity:** coronal, mid-shaft, peno-scrotal, perineal		self-developed questions		-		-

Table 1 continued

Study references (authors)	Participants (hypospadias group)	Controls	Measurement of four outcome-measures concerning genital self-perception	Outcome: Negative genital perception (measured with a sum score)	Outcome: Dissatisfaction with general penile appearance	Outcome: Concern of abnormal appearance/ Perceived difference in appearance from others	Outcome: Dissatisfaction with penile length
Hoag, Gotto, Morrison, Coleman, and Macneily (2008)	**N:** 28 **Mean-age:** 20.00 years **Age-range:** 16-31 years **Severity:** anterior, middle, posterior		self-developed questions		-		
Jiao, Wu, Xu, and Yu (2011)	**N:** 43 **Mean-age:** 21.6 years **Age-range:** 18.1-34.2 years **Severity:** distal, proximal		self-developed questions		+	+	
Kenawi (1976)	**N:** 82 **Age-range:** 18-35 years **Severity:** glandular, coronal, penile, penoscrotal, scrotal, perineal		self-developed questions		-		-

Table 1 continued

Study references (authors)	Participants (hypospadias group)	Controls	Measurement of four outcome-measures concerning genital self-perception	Outcome: Negative genital perception (measured with a sum score)	Outcome: Dissatisfaction with general penile appearance	Outcome: Concern of abnormal appearance/ Perceived difference in appearance from others	Outcome: Dissatisfaction with penile length
Kiss et al. (2011)	**N:** 104 **Mean-age:** 31.6 +/-6.0 years **Age-range:** 24-42 years **Severity:** midshaft, proximal	**N:** 63 age-matched healthy men without genital malformation **Mean-age:** 32.7 +/- 4.7 years	self-developed questions		**+c**		
Kumar and Harris (1994)	**N:** 35 **Mean-age:** 16 years **Age-range:** 13-25 years **Severity:** posterior		self-developed questions			-	-
Liu et al. (2006)	**N:** 102 **Mean-age:** 24.5 years **Age-range:** 19.2-33.9 years **Severity:** distal, proximal		self-developed questions		-		

Table 1 continued

Study references (authors)	Participants (hypospadias group)	Controls	Measurement of four outcome-measures concerning genital self-perception	Outcome: Negative genital perception (measured with a sum score)	Outcome: Dissatisfaction with general penile appearance	Outcome: Concern of abnormal appearance/ Perceived difference in appearance from others	Outcome: Dissatisfaction with penile length
Mondaini, Ponchietti, Bonafè, et al. (2002)	**N:** 42 **Age:** 18 years **Severity:** anterior, other	**N:** 500 men; randomly selected from 11'649 men undertaking compulsory National service	self-developed questions		**+c**		
Mor, Ramon, and Jonas (2000)	**N:** 43 **Mean-age:** 21.5 years **Age-range:** 16-31 years **Severity:** Distal		self-developed questions			**-**	
Moriya et al. (2006)	**N:** 22 **Mean-age:** 20.6 +/- 2.1 years **Severity:** mild, severe, unknown	**N:** 38 age matched, normal male volunteers **Mean-age:** 21.0 +/- 1.9 years	self-developed questions		**=c**	**=c**	

Table 1 continued

Study references (authors)	**Participants (hypospadias group)**	**Controls**	**Measurement of four outcome-measures concerning genital self-perception**	**Outcome: Negative genital perception (measured with a sum score)**	**Outcome: Dissatisfaction with general penile appearance**	**Outcome: Concern of abnormal appearance/ Perceived difference in appearance from others**	**Outcome: Dissatisfaction with penile length**
Mureau, Slijper, van der Meulen, et al. (1995)	**N:** 73 **Mean-age:** 25.6 +/- 5.7 years **Age-range:** 18-38 years **Severity:** distal, proximal	**N:** 50 men treated for an inguinal hernia **Mean-age:** 25.3 +/- 3.7 years **Age-range:** 18-38 years	Genital appraisal score	**+c**	**=c**	**+c**	**+c** (considering flaccid penis to be too small) **=c** (considering erected penile size to be too small)
Mureau, Slijper, Slob, et al. (1995)	**N:** 73 **Mean-age:** 25.6 +/- 5.7 years **Age-range:** 18-38 years **Severity:** distal, proximal	**N:** 50 men treated for an inguinal hernia **Mean-age:** 25.3 +/- 3.7 years **Age-range:** 18-38 years	GPS	**+c**	**+c**	+	
Olofsson, Oldbring, Becker, Aberg, and Svensson (2003)	**N:** 21 **Age-range:** 20-30 years **Severity: -**		Semi-structured interview with self-developed questions		**-**		

Table 1 continued

Study references (authors)	Participants (hypospadias group)	Controls	Measurement of four outcome-measures concerning genital self-perception	Outcome: Negative genital perception (measured with a sum score)	Outcome: Dissatisfaction with general penile appearance	Outcome: Concern of abnormal appearance/ Perceived difference in appearance from others	Outcome: Dissatisfaction with penile length
Rynja et al. (2009)	**N:** 66 **Mean-age:** 22.0 +/-3.55 years **Severity:** anterior, midshaft, posterior	**N:** 151 medical students **Mean-age:** 21.8+/- 3.0 years; **Age-range:** 18-30 years	self-developed questions PPS		**-**		**+c**
Sommerlad (1975)	**N:** 60 **Age:** ≥ 17 years **Severity:** middle third of the penis or more proximal; distal third; coronal		self-developed interview			+	
Wang et al. (2009) Wang et al. (2010)	**N:** 130 **Medan-age:** 26.19 years **Severity:** distal, proximal	**N:** 50 healthy adult males **Median-age:** 26.2 years **Age-range:** 24-35 years	self-developed questions		**+c**		

Table 1 continued

Study references (authors)	Participants (hypospadias group)	Controls	Measurement of four outcome-measures concerning genital self-perception	Outcome: Negative genital perception (measured with a sum score)	Outcome: Dissatisfaction with general penile appearance	Outcome: Concern of abnormal appearance/ Perceived difference in appearance from others	Outcome: Dissatisfaction with penile length
Weber et al. (2013)	**N:** 15 **Mean- Age:** 25.4 years **Age-range:** 19-39 years **Severity:** distal & proximal		PPS	-			

Notes:

+c Indicates that **more men with corrected hypospadias** compared to controls have a negative genital self-perception, are disatisfied with their genital appearance, are concerned about abnormal penile appearance or feel different from other men, or are dissatisfied with their penile length

-c Indicates that **fewer men with corrected hypospadias** compared to controls have a negative genital self-perception, are dissatisfied with their genital appearance, are concerned about abnormal penile appearance or feel different from other men, or are dissatisfied with their penile length

= c Indicates that **men with corrected hypospadias do not differ** from controls regarding genital self-perception, satisfaction with genital appearance, evaluation of normality about penile appearance or feelings of being different from other men, or satisfaction with their penile length

+ Indicates that **most men with corrected hypospadias** have a negative genital self-perception, are dissatisfied with their genital appearance, are concerned about abnormal penile appearance or feel different from other men, or are dissatisfied with their penile length

- Indicates that **less than half of men or only a few men with corrected hypospadias** have a negative genital self-perception, are dissatisfied with their genital appearance, are concerned about abnormal penile appearance or feel different from other men, or are dissatisfied with their penile length

2.3.2. Genital self-perception of men with corrected hypospadias explained with the Cognitive-Behavioral (CB) model of body image by Cash

As mentioned in chapter A 2.1., the term "body cathexis" was defined by Secord and Jourard (1953, p. 343) as "the degree of feeling of satisfaction or dissatisfaction with the various parts and processes of the body". Genital self-perception can therefore be regarded as one aspect of the entire body perception.

Theoretical models relating to the development of genital self-perception of hypospadias patients are lacking. However, there is a well accepted model in the field of body image: Cash's Cognitive-Behavioral (CB) perspectives (Cash, 2002, 2011), which will be described in more detail in the following chapter. As discussed above, genital self-perception may be regarded as one aspect of the entire body image. Therefore, the CB model is applied in this dissertation to explain the development of genital self-perception for men with corrected hypospadias.

Cash's CB model exhibits several advantages. First, it can be applied not only to unaffected individuals but also to those with a malformation or a disfigurement (Rumsey & Harcourt, 2005, Chapter 2) such as hypospadias. Second, it can explain both the development of positive and negative body images (Cash, 2002, 2011; Rumsey & Harcourt, 2005, Chapter 2) and genital self-perception. Third, it provides an overview of the most relevant factors that may influence the development of a positive or negative body image and genital self-perception. It thus helps to identify the gaps and limitations in the current literature which are relevant for the generation of new research questions (Cash, 2002, 2011).

The following chapter (A 2.3.2.1.) explains Cash's CB model (Cash, 2002, 2011). In chapter A 2.3.2.2., the CB model is set in relation to genital self-perception of men with corrected hypospadias.

2.3.2.1. Cognitive-Behavioral (CB) model of body image by Cash

With Cash's Cognitive-Behavioral (CB) perspectives (Cash, 2002, 2011) we have a heuristic conceptual model that represents the multidimensionality of body image. Cash emphasizes that the CB model describes no single theory but rather several theoretical aspects, such as social learning, conditioning processes, and the mediation of behavior and emotion by cognitive processes. Figure 2 illustrates the dimensions, determinants, and processes of Cash's CB model. The central organizing construct of the CB model is *body image attitudes*. It consists of two basic attitudinal elements, *body image evaluations* (appraisal and beliefs about one's appearance) and *body image investments* (individual's cognitive, behavioral, and emotional importance concerning one's physical appearance).

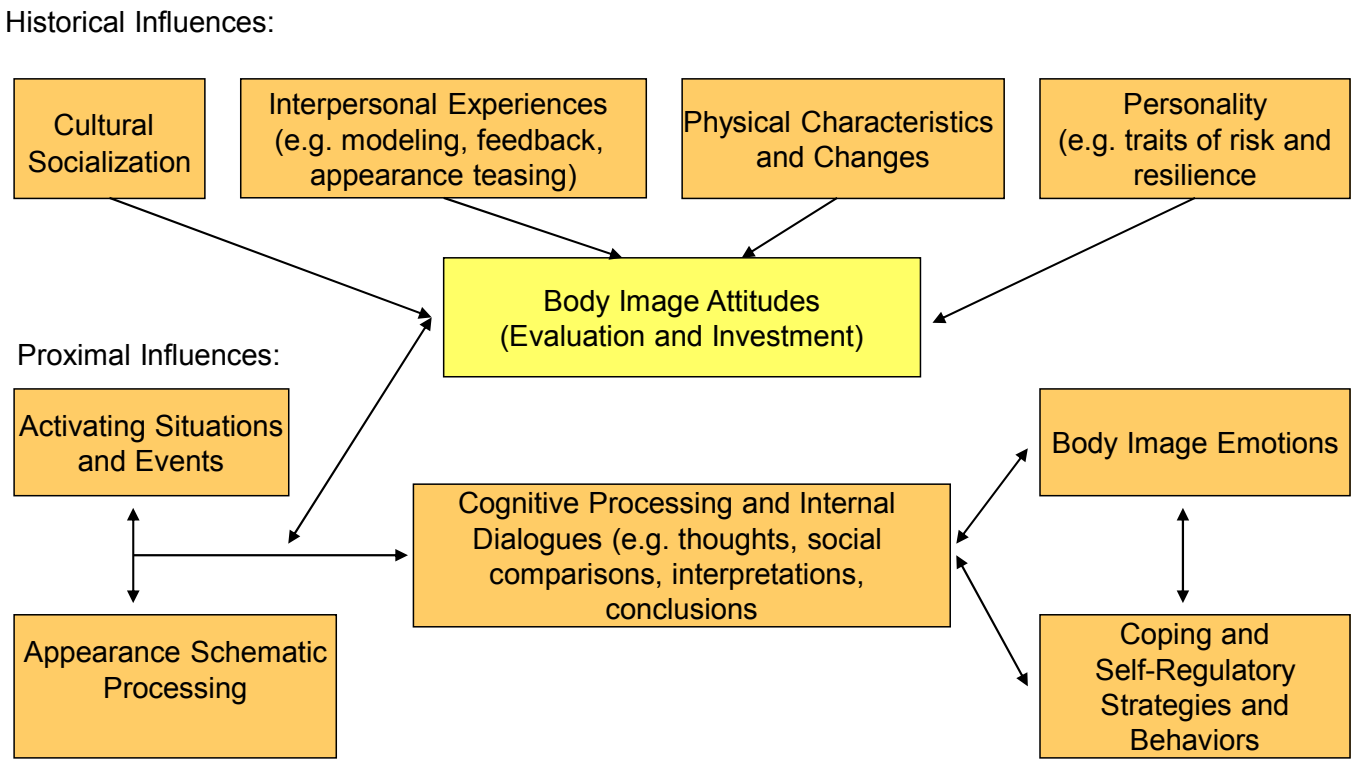

Figure 2: Cognitive - Behavioral (CB) Perspectives on Body image by Cash: dimensions, determinants, and processes according to Cash (2011, p. 41)

In addition, body image attitudes include *self-schemas* in relation to one's appearance (a higher-order construct that expresses one's body image investment).

According to the CB model, body image attitudes are shaped by past, historical influences and proximal or concurrent factors. Historical influences concern experiences during childhood and adolescence in relation to one's own physical appearance and socialization about its meaning. Four historical influences are important for the development and function of body image attitudes, including self-schemas. These influences are cultural socialization, interpersonal experiences, physical characteristics and changes, and personality factors.

Important present influences within the CB model are activating situations and events, cognitive processing and internal dialogues, body image emotions, and coping, respectively self-regulatory strategies and behaviors.

The distinction between past and present influences makes the model highly dynamic, as present influences become historical influences in the future (Cash, 2002, 2011; Rumsey & Harcourt, 2005, Chapter 2). Although the illustration of the CB model contains arrows, Cash himself (Cash, 2011) pointed out that causality is more complex. He refers to Albert Bandura's social cognitive theory of triadic reciprocal causation (Bandura, 1986). This theory states that within an individual there is a mutual influence between all variables (behavior, personality and environment). Therefore, Cash emphasizes that all variables of the CB model may interact with each other.

The model is explained in more detail in the following sections.

Historical influences

Cultural socialization

Every culture and subculture owns and transmits, for instance through media, standards or expectations about what is seen as attractive or unattractive and about which attributes are typically female and male. Individuals internalize those cultural messages and norms which influence their basic body image attitudes (Cash, 2002, 2011).

Interpersonal experiences

Individuals also learn about appearance values, attitudes and behaviors through observation and interaction with their family, friends, peers, and even strangers. There are different ways in which appearance-related information is transmitted. First, family members or individuals

outside the family can convey direct messages (e.g. by teasing or making comments about a child's appearance). The interpretation of these direct messages is influenced by additional cues like tone of voice, context and the supposed motives of the speaking person. Second, appearance-related information can be transmitted indirectly by role modeling when children are exposed to parents' and siblings' appearance-related attitudes and behaviors. These direct and indirect messages provide a standard against which a child compares him- or herself (Bellew, 2012; Cash, 2002, 2011).

Physical characteristics and changes

The development of body image is not only affected by such environmental factors as social feedback on appearance. It is also influenced by an individual's actual physical characteristics. Cash (2002) refers to the goodness-of-fit-model by Lerner and Jovanovic (1990), which states that how well one's physical appearance fits social standards of attractiveness is crucial for self-appraisal. In this process, social feedback (e.g. rejection or teasing) acts as a mediator. Of course, satisfaction with one's own physical appearance is also influenced by self-evaluations in relation to internalized standards (Cash, 2002, 2011).

During lifetime, the human body changes in relation to physical competence and physical appearance. For example, dramatic changes happen during pubertal maturation, through operations and with acquired disfigurements. Thus, Cash (2011, p. 42) concludes that "the body is a 'moving target', which entails an ongoing process of adaption to physical changes, for better or for worse."

Personality Factors

Other important influences that contribute to the development of body image are personality traits. Some of them represent risk factors for the development of a negative body image and some of them promote a positive body image. In the CB model, Cash (2002, 2011) gives an overview of the most important personality traits that may act as risk factors or foster resilience against the development of a negative body image. First, high self-esteem may not only promote the development of a positive body image but also act as a buffer in situations in which one's body image is threatened. Second, perfectionism may facilitate the pursuit of high ideals of physical appearance that can hardly be achieved or even not at all. This may result in body dissatisfaction. Third, a secure attachment may determine a positive body appraisal, while an insecure attachment may lead to a dysfunctional body image. Furthermore,

public self-consciousness, a need for social approval and gender-role attitudes may also influence one's body evaluation.

Proximal influences

Body image attitudes including self-schemas, activating events and cognitive Processing

As already mentioned, historical variables influence the development of certain body image attitudes. As illustrated in the CB model (Figure 2), (Cash, 2011, p. 43) "body image attitudes are central organizing constructs in the interplay of cognitive, emotional, and behavioral processes that occur within the context of environmental events". They are predispositions by which individuals interpret and react to their own physical appearance and to appearance-related events. Besides self-schemas, body image attitudes contain two basic attitudinal elements, body image investment and body image evaluation. Thomas Cash defines the two components of body image as follows (Cash, 2011, p. 39): "Body image evaluation refers to individuals' satisfaction or dissatisfaction with their body and their evaluative beliefs about it. Body image investment refers to the cognitive, behavioral, and emotional importance of the body for self-evaluation."

Self-schemas help individuals to interpret information they perceive in their daily life. Markus (1977, p. 64) defined the term self-schema as "cognitive generalizations about the self, derived from past experience, that organize and guide the processing of self-related information contained in an individual's social experience". For example, individuals that are appearance-schematic pay more attention to information in relation to their appearance and have a specific schema for this particular self-dimension. In contrary, individuals that are appearance-aschematic are not concerned about their appearance because they do not have a particular appearance schema. Therefore, appearance-schematic individuals process any appearance-related information differently (i.e., appropriately to their schema) than appearance-aschematic individuals. There are specific events or situational cues (e.g. seeing one's reflection in the mirror) that trigger schema-driven processing of appearance-related information. In addition, internal dialogues (automatic thoughts and conclusions about one's physical appearance) are activated. These processes may be biased, as for instance is the case in anorexic women.

Body image emotions, coping and self-regulatory strategies and behaviors

An individual has several *body image emotions* in relation to these processes. An individual develops several cognitive or behavioral strategies to cope with negative anticipated or real

emotions and thoughts. Cash (2002, p. 43) identified three ways to cope with distressing body image thoughts or emotions. First, *Avoidance*: "Attempts to avoid the threat to one's thoughts and feelings about body image". Second, *Appearance Fixing:* "Attempts to change one's appearance by concealing, camouflaging, or 'fixing' a physical characteristic perceived as disturbing". Third, *Rational Positive Acceptance:* "Actions or strategies that focus on positive self-care or rational self-talk and acceptance." Cash's study results (Cash, 2002) indicated that avoiding specific situations, thoughts or feelings as well as fixing a disturbing part of the body are significantly associated with dysfunctional body image schemas. In contrast, rational coping was not found to correlate with these negative schemas. The author concluded that the use of poor coping strategies possibly leads to increasing negative self-appraisals and to the perpetuation of body image distress.

In the following section (A 2.3.2.2.), the CB model is set in relation to genital self-perception of men with corrected hypospadias.

2.3.2.2. Genital self-perception of men with corrected hypospadias explained with the Cognitive-Behavioral (CB) model of body image by Cash

As previously mentioned, genital self-perception can be regarded as one aspect of the entire body image. Therefore, in this chapter, all the dimensions, determinants, and processes of the CB model discussed above are set in relation to the genital self-perception of men with corrected hypospadias. Specifically, this section describes study results and theoretical reflections concerning the four historical factors and the proximal variables that may influence the development and maintenance of a positive or negative genital self-perception.

Cultural Socialization in relation to genital self-perception of men with corrected hypospadias

Until now, no studies have directly investigated the influence of norms such as those delivered by media (such as pornography) on the genital self-perception of hypospadias patients. However, some results indirectly reflect some influences of cultural norms. In European countries, most men are not circumcised. In line with this cultural norm, some European studies among men with corrected hypospadias (Aulagne et al., 2010; Mureau, Slijper, van der Meulen, et al., 1995; Mureau, Slijper, Slob, et al., 1995) indicated that the circumcised appearance of the penis after hypospadias repair correlates with negative genital self-perception. In contrast, some Turkish authors (Eray et al., 2005) have suggested that the

tradition of circumcision in their country, where circumcision is very common for religious reasons, means hypospadias patients have no negative effect on their body image after hypospadias surgery.

Interpersonal experiences in relation to genital self-perception of men with corrected hypospadias

Many authors believe that interpersonal experiences in relation to hypospadias may have a profound influence on a child. For example, both positive and negative feelings and attitudes of parents towards their child's hypospadias (e.g. worries in relation to their son's sexuality) might be transmitted to their son. The emotional reaction of parents is therefore crucial for children's feelings toward their hypospadias and thus for their genital self-perception (e.g. Denson & Terry, 1988; Easson, 1966). Some authors even assume that the emotional reaction of a hypospadias patient's environment (e.g. family and society) "may be even more crippling to his total emotional and physical growth than the physical defect itself" (Easson, 1966, p. 453).

As with other congenital malformations, the presence of hypospadias may cause feelings of guilt in both parents and in affected children. In addition, the fact that a hypospadias is not usually visible to others favors the creation of a "family secret". Affected children may interpret the secrecy as an indication that much is wrong with them (Robertson & Walker, 1975).

There are few studies regarding reactions of the social environment towards hypospadias (e.g. ridicule or rejections of peers or potential sexual partners) (e.g. Mureau, Slijper, Nijman, et al., 1995; Vandendriessche et al., 2010). Those studies will be described in more detail in chapter A 2.4.1. Studies that have investigated the relationship between patients' genital self-perception and reactions of the social environment towards patients' penile appearance have clearly shown that receiving negative comments by peers or experiences of being ridiculed by partners correlated with a worse genital self-perception of hypospadias patients (e.g. Liu et al., 2006; Mureau, Slijper, Slob, et al., 1995). In contrast, receiving information about hypospadias (e.g. the reason for having been operated) were found to be positively associated with a more positive genital self-perception of hypospadias patients (Schonbucher et al., 2008b).

Physical characteristics and changes in relation to genital self-perception of men with corrected hypospadias

Long-term studies on hypospadias repair often investigate the interaction between the type of hypospadias and patients' genital self-perception. Most studies have shown that men with a proximal hypospadias reported being more dissatisfied with their penile appearance (e.g. Chertin et al., 2013; Liu et al., 2006; Mureau, Slijper, van der Meulen, et al., 1995; Mureau, Slijper, Slob, et al., 1995; Rynja, de Jong, et al., 2011; Rynja, de Kort, & de Jong, 2012) and their penile size (e.g. Berg et al., 1981; Mureau, Slijper, van der Meulen, et al., 1995; Rynja, de Jong, et al., 2011) than men with distal hypospadias. In addition, more men with proximal hypospadias considered their penises to look different from those of other men than men with distal hypospadias (e.g. Jiao et al., 2011). However, a few studies found no significant correlation between type of hypospadias and satisfaction with penile appearance (e.g. Aho et al., 2000; Weber et al., 2013).

Next to physical characteristics of hypospadias, long-term studies on hypospadias repair often examine the correlation between genital self-perception and other variables such as medical factors, patients' sexuality and patients' demographic variables. These variables are also related to physical characteristics (e.g. erectile function) or physical changes (e.g. surgical techniques & complications) of men with corrected hypospadias and are therefore described in this section.

Regarding medical factors, some research has examined whether the type of surgical technique influences patients' genital self-perception. Some authors (e.g. Aho et al., 2000; Aho et al., 1997; Mureau, Slijper, van der Meulen, et al., 1995; Mureau, Slijper, Slob, et al., 1995) found no correlation between surgical technique and patients' satisfaction with their penile appearance. In contrast, other authors (e.g. Liu et al., 2006) found that more men who had been operated by two-stage techniques than those operated by one-stage techniques reported being dissatisfied with their genital appearance. As nowadays hundreds of reconstructive techniques are available (Baskin & Ebbers, 2006), though, these results do not allow general conclusions regarding the influence of surgical techniques on hypospadias patients' genital self-perception. Other medical factors investigated are surgery-related complications and the number of operations. Some studies found a correlation between the complication rate (e.g. Liu et al., 2006) or the number of operations (e.g. Liu et al., 2006; Mureau, Slijper, Slob, et al., 1995) and genital self-perception. In contrast, other studies found no such association (e.g. Bubanj et al., 2004; Mondaini, Ponchietti, Bonafè, et al., 2002; Mureau, Slijper, van der Meulen, et al., 1995; Weber et al., 2013). Inconsistent results also

arise in the relation between genital self-perception and the age at which surgery was completed. Whereas some studies found a significant negative correlation (e.g. Wang et al., 2010) or a negative trend (e.g. Mureau, Slijper, Slob, et al., 1995) between age at last operation and genital self-perception, other investigations found no such correlation (e.g. Bubanj et al., 2004; Mondaini, Ponchietti, Bonafè, et al., 2002; Weber, Schonbucher, Gobet, Gerber, & Landolt, 2009).

Regarding patients' sexuality, results indicated that men who are satisfied with the appearance of their HASRGs have fewer problems with erection and ejaculation and more reported being sexually experienced than unsatisfied patients (Jiao et al., 2011). However, Nelson, Bloom, Kinast, Wei, and Park (2005b) found that satisfaction with penile appearance was neither significantly associated with the IIEF-scores nor with overall sexual satisfaction.

Regarding satisfaction with penile length, study results showed that it seems to correlate positively with relationship quality (Rynja et al., 2009).

Only a few studies have examined the possible relation of demographic variables and genital self-perception in men with corrected hypospadias. They showed that higher age (in children and adolescents) (Mureau, Slijper, Nijman, et al., 1995; Weber et al., 2008) and higher education (in adults) (Liu et al., 2006) correlated with a lower satisfaction with penile appearance.

Personality in relation to genital self-perception of men with corrected hypospadias

To our knowledge, there is no study that evaluates personality traits in relation to genital self-perception of men with corrected hypospadias.

Proximal influences

According to Cash's CB model (Cash, 2002, 2011), genital self-perception of men with corrected hypospadias is shaped by historical influences and by proximal or concurrent determinants and processes. Only a few studies have examined the cognitive, emotional and behavioral processes that interact with genital self-perception. Their results show that men with hypospadias seem to suffer more often from sexual inhibition than controls. They are more often inhibited in seeking sexual contact (e.g. Mondaini, Ponchietti, Bonafè, et al., 2002;

Mureau, Slijper, van der Meulen, et al., 1995; Wang et al., 2010) or are more often afraid of being ridiculed by others because of the appearance of their penis (e.g. Wang et al., 2010).

Another research team published results concerning coping styles of men with corrected hypospadias in a congress abstract (Rynja, De Kort, Bosch, & Kok, 2011). Interestingly, of seven possible coping styles, avoidance was more often applied by men with corrected hypospadias than a reference group in the Dutch population. In line with Cash's study results (Cash, 2002), the avoidance coping style was significantly associated with a lower satisfaction with penile appearance and body image.

After discussing genital self-perception of men with corrected hypospadias, the next chapter (chapter 2.4.) provides an overview of the current literature concerning genital perception of HASRGs by others.

2.4. Perception of hypospadias-affected surgically-repaired genitals by others

2.4.1. Research on perception of hypospadias-affected surgically-repaired genitals by others

As discussed above, men with corrected hypospadias seem to be concerned about negative reactions such as ridicule of peers or of a potential sexual partner because of their penile appearance (e.g. Mondaini, Ponchietti, Bonafè, et al., 2002; Mureau, Slijper, van der Meulen, et al., 1995; Wang et al., 2010). Only a few studies have investigated the reactions of the social environment such as peers or sexual partners. Whereas older studies found that more hypospadias patients than controls had received negative comments (Mureau, Slijper, Nijman, et al., 1995) or had been bullied (Svensson, Berg, & Berg, 1981) because of their penile appearance, a more recent one (Vandendriessche et al., 2010) showed that adolescents with operated hypospadias were not ridiculed significantly more often than non-affected controls. However, these results only reflect the patients' views. Analysis of the situation of men with corrected hypospadias based on Cash's CB model (Cash, 2002, 2011) may lead to the prediction that those with a negative genital self-perception tend to interpret any comment in relation to their penis in a negative way, regardless of whether the comment was formulated neutrally or was actually negative. Thus, it would be interesting to know how HASRGs are actually perceived by others.

Interestingly, thus far, no investigation has assessed how laypersons unacquainted with hypospadias such as peers evaluate HASRGs. The only studies about genital perception of HASRGs by others and not by the patients themselves have investigated the evaluation of

HASRGs by medical personnel, parents and intimately involved persons (e.g. Baskin, 2001; Mureau, Slijper, Slob, Verhulst, & Nijman, 1996; Scarpa, Castagnetti, Musi, & Rigamonti, 2009; Snodgrass, Ziada, Yucel, & Gupta, 2008; Ververidis, Dickson, & Gough, 2005; Weber et al., 2013; Weber et al., 2008). One of these studies (Weber et al., 2013) examined urologists' genital perception of adult men with corrected hypospadias, whereas most of them examined urologists' genital perception of children or adolescents (e.g. Baskin, 2001; Mureau et al., 1996; Scarpa et al., 2009; Snodgrass et al., 2008; Ververidis et al., 2005; Weber et al., 2008). This study with adults was conducted by our research team in the context of this doctoral thesis. Results showed that men with corrected hypospadias rated their own genitals similarly to a group of independent urologists. Interestingly, in an recent study by the same primary author (Weber et al., 2008), children and adolescents with corrected hypospadias reported a higher genital self-perception than independent urologists. The authors (Weber et al., 2013) suggested that, compared to children and adolescents, adults rate more like urologists because of higher expectations and a more critical self-evaluation.

Although studies regarding genital perception by individuals without any knowledge of hypospadias (e.g. peers and sexual partners) are lacking, there is one study (Maier & Tewes, 1984) which indicates that the cosmetic penile appearance of men with corrected hypospadias in many case does not bother their intimate partners. Maier and Tewes (1984) noted that, in their interviews on sexuality, no partner of men with corrected hypospadias criticized their partner's penile appearance.

2.4.2. Factors that influences the perception of hypospadias-affected surgically-repaired genitals by others

The study mentioned above on genital perception of adult HASRGs by others (Weber et al., 2013) also produced the notable finding that urologists did not fully agree in their evaluation of the penile appearance of several photographs of HASRGs. For example, while some urologists were "very satisfied" with the general cosmetic penile appearance of an illustrated penis, others evaluated the appearance only as "satisfactory" or even as "not satisfactory". The low interrater reliability (ICC = .46) probably indicates that both characteristics of the illustrated penis (e.g. severity of hypospadias) and variables of the rating perceivers (e.g. rater's gender and age) influence their penile perception of others. However, this study did not investigate whether any perceiver-related variables can be used to predict the evaluation of HASRGs. Instead, the influence of hypospadias-related characteristics, the severity of hypospadias and the number of operations, was examined. Importantly, although these characteristics correlated with urologists' evaluations of HASRGs, they did not correlate with patients' genital self-perception. Thus, urologists' perception of HASRGs seems to be more likely influenced by objective criteria than the patients' genital self-perception.

Studies in children and adolescents with corrected hypospadias revealed a few variables that seem to correlate with raters' perception of HASRGs. First, intimately involved persons such as parents were found to rate differently than medical personnel such as surgeons (Snodgrass et al., 2008; Weber et al., 2008). However, other findings show good agreement between parents' and surgeons' ratings of HASRGs (Holland et al., 2001; Scarpa et al., 2009). Second, one set of results indicated that independent surgeons rated more critically than affiliated ones (Weber et al., 2013). Third, Weber et al. (2008) speculated in their study that feelings of fear and guilt in relation to penile malformation possibly influenced parents' evaluations of their sons' HASRGs.

2.5. Chapter summary

In summary, there are only a few studies regarding genital self-perception of men with corrected hypospadias and even fewer concerning genital perception of HASRGs by others. A main weakness of the studies is their lack of reference to psychological theories and their use of self-developed, unvalidated questionnaires.

However, the findings on genital self-perception clearly demonstrate that men with corrected hypospadias have a significantly worse genital self-perception than men without hypospadias.

Since there is no specific theory regarding genital self-perception of individuals with corrected hypospadias or of men without any genital malformation, Cash's CB model of body image is used to explain which factors may influence the development of hypospadias patients' genital self-perception. Studies involving the factors that may influence the development of genital self-perception have mainly investigated the influence of medical and hypospadias-related variables; no study has examined the influence of personality traits. Results concerning hypospadias-related characteristics clearly showed that more severe types of hypospadias are associated with worse genital self-perception. In contrast, findings on medical variables (e.g. number of complications and age at operation) are inconsistent. This may be due to methodological limitations (c.f. p.36). Regarding socio-cultural influences, some findings indicate that some kinds of norm regarding penile appearance may influence patients' genital self-perception (e.g. norms regarding a circumcised penis). However, it still remains unclear how a penis has to look to be perceived as good or normal by the general population. According to Cash and to several other authors, interpersonal influences such as feelings and attitudes of the social environment towards hypospadias are of crucial

importance for the development of patients' genital self-perception. However, very few studies actually examine the reactions of others in relation to the corrected hypospadias. Those that do show that negative reactions by others are associated with a negative genital self-perception of hypospadias patients.

Studies regarding genital perception by others are scarce and they mainly investigate the evaluation of HASRGs by medical personnel, parents and intimately involved persons. Thus, it remains unclear how individuals without any knowledge of hypospadias would evaluate HASRGs.

3. Health-related quality of life in men living with hypospadias

3.1. Health-related quality of life

3.1.1. The concept of health-related quality of life

Due to improvements in medical technique, some lethal illnesses have become chronic diseases during the last century. Thus, survival time is no longer the only criterion to describe the effectiveness of a therapy. Besides physical variables, psychological and social factors are also variables relevant to the outcome assessment of a medical therapy. In this context, health-related quality of life (HRQoL) is an important concept. In contrast to quality of life (QoL), which is a broader concept, health-related quality of life (HRQoL) only refers to human experience and behavior in relation to health and disease (Landolt & Sennhauser, 2007).

HRQoL is defined as "a multidimensional concept that includes the broad areas of functional status, psychologic and social well-being, health perceptions, and disease- and treatment-related symptoms" (Aaronson et al., 1991, p. 840). There are three dimensions that cross-culturally represent the core of HRQoL (see Figure 3): first, physical functions such as physical symptoms and complaints; second, psychological functions such as the emotional state and the cognitive functioning; and third, social functions such as the quality of relations with family members and peers. The concept of HRQoL is highly dynamic and changes, for example, with the course of the disease and with changing life circumstances (Landolt & Sennhauser, 2007).

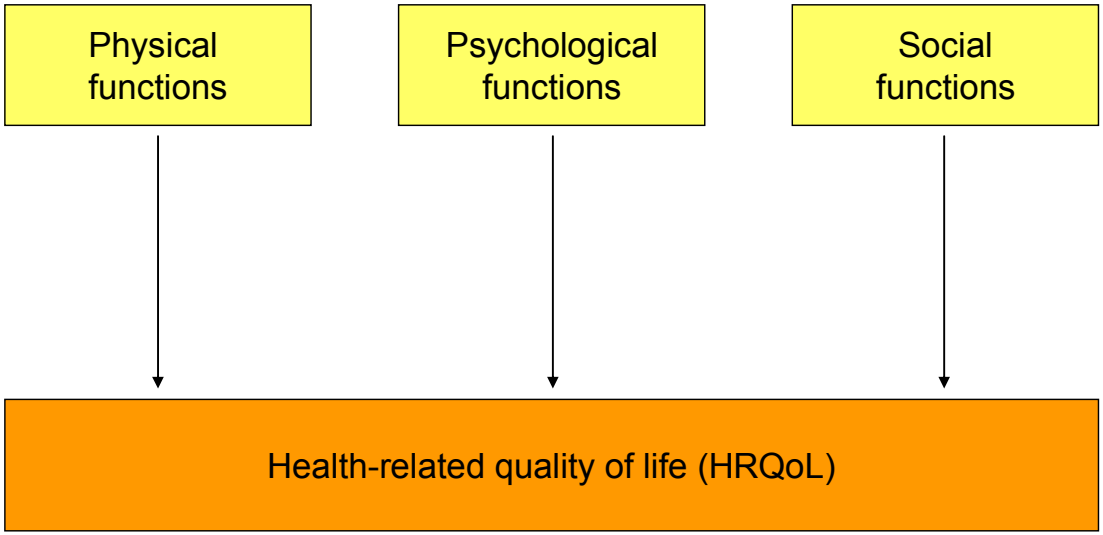

Figure 3: Dimensions of health-related quality of life according to Landolt and Sennhauser (2007, p. 125)

Individuals' self-report is the standard way to measure HRQoL. However, proxies (such as parents or medical personnel) may also provide information on an individual's HRQoL to offer a complementary perspective. Furthermore, proxy ratings are needed when an individual is not able to give information regarding his or her HRQoL (e.g. in children when necessary language skills are lacking) (Landolt & Sennhauser, 2007; Levine, 1995).

There are different categories of QoL measures. Generic measures such as the Medical Outcomes Study Short Form-36 item questionnaire (SF-36) (Ware, Snow, & Kosinski, 2000)

have been developed to assess the HRQoL of patients and of the general population. Their advantage is that patient groups can be compared with one another or with healthy populations. However, they might not be sensitive enough to measure concerns that are indicative of a condition. In contrast, disease- or population-specific QoL measures focus on the particular problems of a certain patient group, but they do not allow comparisons with the general population. Therefore, both types of measures are applied in many studies (McGee & Ring, 2010).

Interestingly, not all ill individuals (even severely ill ones) have an impaired HRQoL. The differences in HRQoL can hardly be explained only by medical parameters. There are several possible alternative factors, for example, expectations of how life should be, comparisons with the well-being of relatives or friends, and the social context (availability of friends and social support). A further important determinant of the quality of life is the cultural context. Every culture responds differently to an affected individual. Despite what one might expect, lepers, for example, are not feared and stigmatized in all societies (Levine, 1995). Furthermore, studies of children who have had an accident or have been chronically ill show that not all of them (even severely ill ones) have an impaired HRQoL. Results show that social parameters such as the quality of the family relationship are more likely to explain such differences in HRQoL than medical parameters (Landolt & Sennhauser, 2007).

3.1.2. Research on health-related quality of life of men with corrected hypospadias

So far, only a few studies have investigated how much the well-being of men with corrected hypospadias is affected by the repaired malformation. However, most of them used self-developed, unvalidated questionnaires (e.g. Aulagne et al., 2010; Olofsson et al., 2003). Only two studies used validated measures to assess the HRQoL of adults with corrected hypospadias. However, in neither study were the results compared with a control group.

The older of these two studies (Miller & Grant, 1997) examined the HRQoL of men with severe types of hypospadias and genital ambiguity (Miller & Grant, 1997). It showed that the majority of men classified their general health as quite good although some reported that their

quality of life and their psychological well-being were impaired because of their urological condition. In a study by Chertin et al. (2013), men with corrected hypospadias reported a physical and mental HRQoL that was within the normal range but with a trend towards lower scores. Compared to their physical HRQoL, men with corrected hypospadias had a lower mental HRQoL.

Other studies have examined the HRQoL of children and adolescents with corrected hypospadias. Jones et al. (2009), who examined adolescents with corrected hypospadias (13-15 years old), found a quality of life that was comparable to published data on "normal" children. In contrast, a study by Schonbucher, Landolt, Gobet, and Weber (2008a) showed that children and adolescents with corrected hypospadias (7-17 years) reported a significantly lower HRQoL in most dimensions than an age-matched control group of boys after hernia repair. However, the older the patients were, the better their HRQoL. Notably, mothers of boys with hypospadias evaluated their sons' HRQoL as significantly better than the boys themselves. The authors remarked that this is contrary to results of studies with chronic diseases. They suggest that mothers of children with hypospadias psychologically suppress the effect of the hypospadias on their sons' quality of life. In line with other studies (Walker, 1998), they observed that many parents did not want to talk about their sons' hypospadias and kept it a family secret.

3.1.3. Research on determinants of health-related quality of life of men with corrected hypospadias

Only a few of the existing studies regarding HRQoL of individuals with corrected hypospadias also examined variables which potentially correlate with a better quality of life. Interestingly, they showed that medical variables such as the severity of hypospadias (Chertin et al., 2013; Schonbucher et al., 2008a) and the number of surgeries (Schonbucher et al., 2008a) were not associated with patients' HRQoL. Even whether patients' hypospadias was corrected before or after they were 18 months did not have a significant correlation with patients' HRQoL (Weber et al., 2009). However, Schonbucher et al. (2008a) found that higher age within the age-range of 7 to 17 years and "being less ashamed of penile appearance" was a predictor of a better HRQoL. Mothers' evaluation of their children's HRQoL was best predicted by "patients' experience of not having been teased about the penis" followed by a more favorable patients' genital perception. Notably, a better mother-reported HRQoL was also significantly predicted by a more severe type of hypospadias. The authors suggest that individuals with more severe forms may be less able to deny their hypospadias than those with mild types and therefore develop better coping strategies. In addition, medical attendance and familial psychosocial support may be better with severe types.

3.1.4. Chapter summary

Most studies concerning the quality of life in individuals with corrected hypospadias use self-developed, unvalidated measures. The few existing studies which used validated instruments show that children have an impaired HRQoL compared to controls without hypospadias. HRQoL seems to improve in adolescence. And adults reported a HRQoL that was within a normal range. Studies regarding possible predictors of HASRGs revealed *increasing age* and *being less ashamed of penile appearance* to be significant predictors of a better HRQoL. However, these findings are based on the HRQoL of children and adolescents. Severity of hypospadias was not associated with self-reported HRQoL either in children and adolescents or in adults with corrected hypospadias.

4. Gaps and limitations in the current literature

4.1. Gaps and limitations in the current literature regarding genital perception of men with corrected hypospadias

Only a few studies have examined the long-term outcomes of hypospadias repair such as genital self-perception. The findings are somewhat contradictory, possibly due to methodological limitations. For example, the overview of the current literature (Table 1; chapter A 2.3.1.) showed very clearly that most studies used self-developed, unvalidated questions instead of an existing genital perception score, for example by Mureau, Slijper, Slob, et al. (1995). In addition, results are often drawn from investigations with a low response rate, heterogeneous patients and data, and lack a control group (Rynja, de Jong, et al., 2011; Singh, Jayanthi, & Gopalakrishnan, 2008). A further serious limitation of the studies is their lack of theoretical background.

The application of Cash's CB model to explain the development of genital self-perception of men with corrected hypospadias reveals several gaps and limitations in the current literature on hypospadias patients' genital self-perception. Of the four proposed historical factors (which according to the analysis based on the CB model possibly influence the development of genital self-perception) it is mainly the influence of one factor that has been investigated. Specifically, most studies examined how physical and medical characteristics correlate with genital self-perception, but investigations regarding the other proposed influencing factors are sparse. These research gaps leave a number of important questions unanswered. First, it is still unclear which personality traits contribute to the development of a better or a worse genital self-perception. Second, with the exception of the norm to be circumcised or not, it has never been investigated what kind of cultural norms exist in relation to penile appearance of HASRGs and thus influence a patient's genital self-perception. Therefore, we still do not know which aspects of penile appearance actually determine what is regarded as normal or abnormal. However, this knowledge would be important, as mild types of hypospadias are mainly operated for cosmetic reasons. Third, although findings indicate that some individuals with corrected hypospadias are afraid of negative reactions to their penile appearance and although theory states that interpersonal experiences such as being ridiculed because of one's penile appearance are of crucial importance to the development of genital self-perception, we do not know what kind of reaction a hypospadias patients actually ought to expect.

Studies concerning genital perception by others have mainly investigated the evaluation of HASRGs by medical personnel, parents and intimately involved persons. Thus it remains unclear how individuals without any knowledge of hypospadias would evaluate HASRGs. For

example, do peers and sexual partners notice a corrected hypospadias? Do they observe possible scars after surgical correction? Or does a meatus that is not at the tip of the glans (as is the case in non-corrected hypospadias) even bother them not at all? Yet, this knowledge would be important for patient counseling and generally for improving the therapy of individuals with hypospadias.

Finally, with the small number of studies regarding possible perceiver-related characteristics that influence perceivers' evaluation of HASRGs, we have little idea which factors might influence whether HASRGs are perceived positively or negatively.

4.2. Gaps and limitations in the current literature regarding health-related quality of life in men living with hypospadias

Besides studies that use self-developed, unvalidated questionnaires, only two studies have investigated the quality of life of *adults* with corrected hypospadias using validated measures. However, neither study compared results with those of a control group. Furthermore, one of these studies only examined men with severe types of hypospadias and genital ambiguity. Thus, the assessment of HRQoL of adults with corrected, isolated hypospadias by means of a validated questionnaire in comparison with a control group is still lacking. However, such a comparison is important, because it may indicate whether and to what extent adults with corrected, isolated hypospadias differ from the general population in this respect. A significant difference would suggest that repaired hypospadias has an effect on patients' HRQoL. In addition, so far, no predictors of a better HRQoL in *adults* with corrected hypospadias has been identified.

In summary, long-term outcome studies on HRQoL and genital perception of men with corrected hypospadias are rare. However, such studies on the long-term psychological outcome of hypospadias repair are of crucial importance. In particular, most types of hypospadias are corrected at a very early age when a child is not able to decide because it is assumed – without any scientific evidence – that early surgery optimizes patients' psychological development.

5. Research Project

This PhD project is part of a more comprehensive research project on the long-term psychological outcome of men with corrected hypospadias. It was initiated by PD Dr. Daniel M. Weber (Department of Pediatric Surgery) and Prof. Dr. Markus A. Landolt (Department of Psychosomatics and Psychiatry) and was conducted at the University Children's Hospital Zürich between September 2008 and January 2012. In her employment as a research fellow at the Hospital, the author of this doctoral thesis contributed crucially to the study design and to the development of several questionnaires. Under the supervision of the two primary investigators (PD Dr. D. Weber & Prof. Dr. M. Landolt), she conducted the recruitment of participants, the data collection, and the statistical analyses and wrote three articles as a first author. The research was financially supported by the Children's Research Center (CRC) of the University Children's Hospital Zurich and the Wyeth Foundation – Swiss Foundation for the Health of Children and Adolescents.

According to the gaps and limitations of the current literature regarding the long-term psychological outcome of hypospadias repair (see chapter A 4), the PhD project had the aims first to assess the genital perception of HASRGs by laypersons unacquainted with hypospadias (project A) and second to examine the HRQoL of men with corrected hypospadias compared to a control group of non-affected, circumcised men (project B). Both projects are explained in more detail in the following sections.

Figure 4 gives an overview of the research aims of Study A and B.

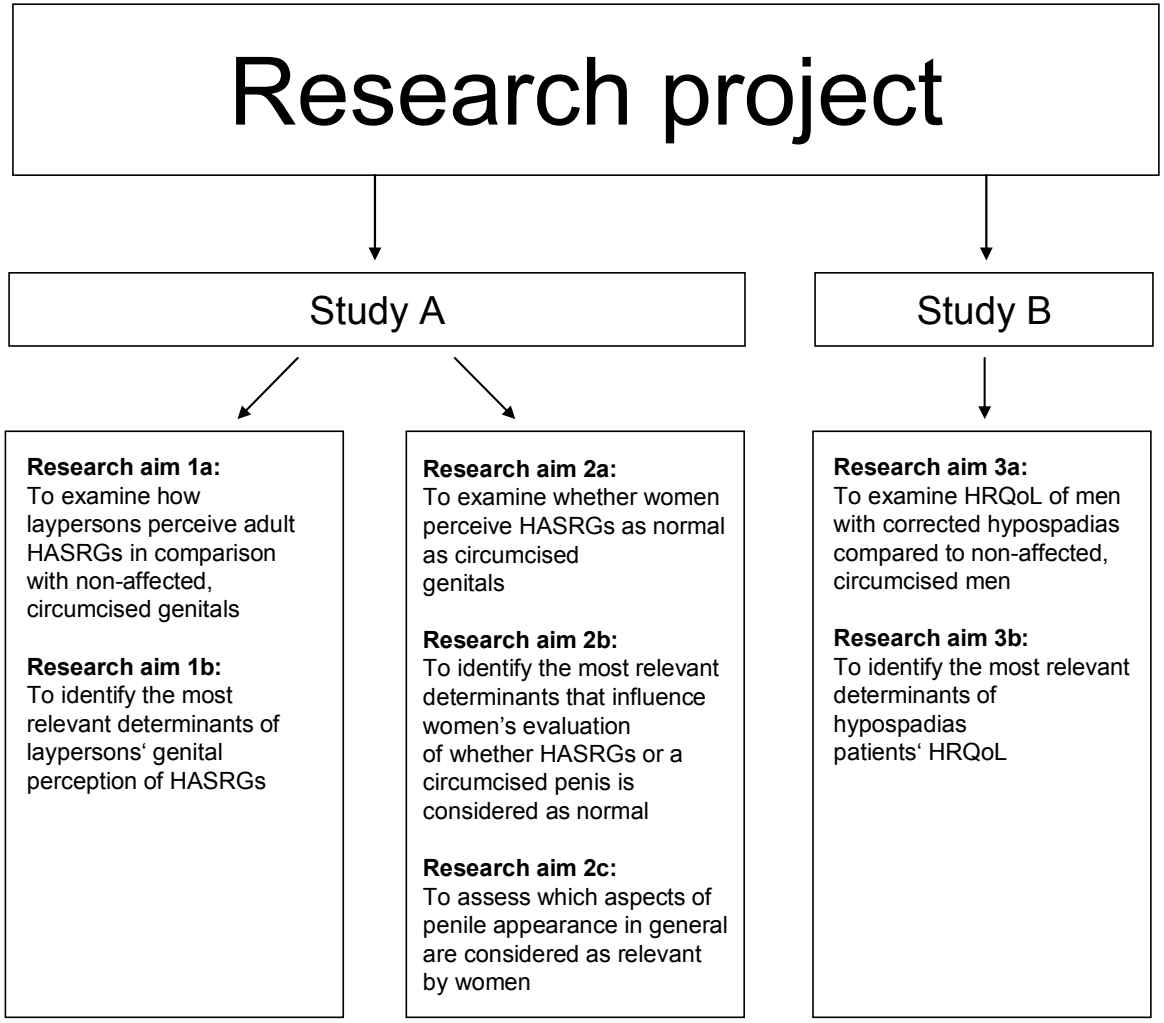

Figure 4: Schematic diagram of the research project and the research aims

5.1. Research project A: Laypersons' genital perception of men with corrected hypospadias and women's rating of the importance of several penile aspects by laypersons

5.1.1. Questions and hypotheses

On reviewing the literature (see chapters A 2.4. & A 4.1.), we found no study which examined how laypersons unacquainted with hypospadias evaluate the genitals of men with corrected hypospadias. However, studies on men with corrected hypospadias clearly showed that some men suffer from sexual inhibition (Mondaini, Ponchietti, Bonafè, et al., 2002; Mureau, Slijper, van der Meulen, et al., 1995; Wang et al., 2010) and are afraid of being ridiculed or rejected by others because of their penile appearance (Wang et al., 2010). In line with Cash's CB model (see chapter A 2.3.2.1.), such concerns may support the development and maintenance of a negative genital perception. Therefore, the knowledge of laypersons' reactions to HASRGs would be of crucial importance for the counseling of hypospadias patients and their parents.

For this reason, we examined the following research questions:

> *Research Question 1a:* Do laypersons unacquainted with hypospadias notice a difference between HASRGs and non-affected, circumcised genitals (controls)?

> *Research Question 1b:* What are perceiver-related characteristics that predict how laypersons evaluate HASRGs?

Because studies on genital perception of HASRGs by laypersons are lacking, our first hypothesis is based on an a priori assumption.

> *Research Hypothesis 1a:* As modern hypospadias repair aims at an anatomical reconstruction of the urethra and usually achieves a good cosmetic result, we hypothesized that laypersons do not notice the difference between HASRGs and circumcised genitals.

Hypothesis 1b is based on the assumption that a deep involvement with sexuality (e.g. with increasing sexual experience at a higher age, with a higher sexual interest or when in an intimate relationship) results in a more favorable genital perception of HASRGs. Moreover, the hypothesis is based on the assumption that knowledge about genital malformations (which someone possibly gains with a higher educational background) has a positive influence on the genital perception of HASRGs by others. Furthermore, it is likely that the way in which an individual evaluates his or her own genital also has an impact on how he or she assesses the genitals of others. Therefore, hypothesis 1b is also based on findings about the genital self-perception of men and women without genital malformations. Those findings indicate that gender and sexual experience have an influence on genital self-perception. Men had a significant more positive genital self-perception

(Morrison, Bearden, Ellis, Harriman, & Morrison, 2004; Reinholtz & Muehlenhard, 1995) and a more positive appraisal of partner's genitals than women (Reinholtz & Muehlenhard, 1995), and sexually inexperienced people rated their genitals significantly worse than experienced ones (Morrison et al., 2004).

> *Research Hypothesis 1b:* Therefore the following variables were expected to be predictors of a more favorable laypersons' perception of HASRGs: male gender of the rater, being in an intimate relationship, higher socio-economic status, higher age, higher sexual interest, higher sexual experience, and a positive genital self-perception of the rater.

In contrast to study 1, which investigated male and female laypersons, study 2 only considered the answers of female laypersons. This restriction was made because most men with corrected hypospadias are heterosexual and thus are interested in the answers of women regarding the research questions of study 2. For example, hypospadias patients are mainly afraid of women's ridicule and rejection in intimate situations. Thus, the opinion of the female laypersons was particularly relevant to the question whether patients' penises were considered to be normal-looking and whether penile length was considered to be a relevant aspect of a penis.

A review of the literature showed that some men with corrected hypospadias perceive their penile appearance to be abnormal (see Table 1, section 2.3.1.). However, it is as yet not clear if only some hypospadias patients or also laypersons consider the penile appearance of

HASRGs as less normal-looking than non-affected genitals. Furthermore, it is unclear which aspects of penile appearance are considered relevant by women, or (following Cash's CB model; see chapter 2.3.2.1.) which cultural norms of penile appearance exist and thus influence whether HASRGs are rated as normal-looking or not. Because mild types of hypospadias (which represent the majority of hypospadias) are mainly surgically repaired for aesthetic reasons or to improve the position and the shape of the meatus, we especially wanted to know how important women consider the meatus in comparison to the other penile aspects. As hypospadias patients are more often dissatisfied with their penile size than non-affected men (Mureau, Slijper, van der Meulen, et al., 1995; Rynja, de Jong, et al., 2011), we were also interested in how relevant women consider penile size to be in comparison to the other penile aspects.

Therefore, our research questions were these:

> *Research Question 2a:* Do women perceive HASRGs as normal-looking as circumcised genitals?

> *Research Question 2b:* Which are the most relevant observer-related predictors for perceiving HASRGs and non-affected, circumcised genitals as normal-looking?

> *Research Question 2c:* Which aspects of penile appearance are considered to be relevant by women?

The following hypothesis is based on the same a priori assumption as hypothesis 1a.

> *Research Hypothesis 2a:* As modern hypospadias repair usually achieves an objectively satisfactory cosmetic result, we hypothesized women to perceive HASRGs as normal-looking as the genitals of non-affected, circumcised men.

Hypothesis 2b is based on the same a priori assumptions and on the same findings as hypothesis 1b. Furthermore, hypothesis 2b is based on Cash's CB model (see chapter

2.3.2.1.). This model postulates that individuals who consider appearance (in our setting penile appearance) as something important process any information regarding penile appearance differently from individuals who regard penile appearance as something unimportant. In line with these theoretical assumptions, we expected that the rating of penile appearance is influenced by whether a woman perceives different penile aspects as important.

> Research Hypothesis 2b: Therefore, we postulated that the following variables were predictors of women's evaluation whether a penis is perceived as normal-looking: higher age, higher degree of sexual interest and a higher number of sexual partners a woman has had during her life. In addition, we expected that the rating of penile appearance is influenced by the fact whether a woman perceives the meatus, the penile size or the general penile appearance as important or not.

Several studies have indicated that most women find penile size "unimportant" or even "totally unimportant" (Francken, van de Wiel, van Driel, & Weijmar Schultz, 2002) and that most women are satisfied with their partner's penis size (Lever, Frederick, & Peplau, 2006). In addition, a study (Fichtner et al., 1995) in "normal" men demonstrated that the position of the meatus varied widely.

> Research Hypothesis 2c: Therefore, we expected that women would consider the position and shape of the meatus and penile size as unimportant penile aspects in comparison to other penile aspects.

5.1.2. Methods

Study design

A cross-sectional study was performed in which a questionnaire with standardized photographs of non-erect HASRGs and circumcised penises was presented to laypersons unacquainted with hypospadias to measure how they rated these genitals.

Subjects (laypersons who completed the questionnaire)

Subjects were individuals without previous knowledge about hypospadias who had no affiliation to the hospital or to any of the patients. They were recruited to three different age groups (age ranges 16-20, 25-30 and 40-45 years); it was assumed that these correlate with different degrees of sexual experience.

The youngest age group (16-20 years) was recruited at schools of two different educational levels. Twenty-five of 305 young men (8.20%) and 33 of 136 young women (24.26%) agreed to participate. Furthermore, two randomly selected population samples of men and women (25-30 years or 40-45 years old) were ordered from a local database agency. Of 800 women and 800 men contacted, 25 women (3.13%) and 38 men (4.75%) were untraceable. Of the remainder, 41 men (5.38%) and 66 women (8.25%), as well as six women and four men who were partners or friends of those invited, agreed to participate.

Study 1 is based on data from the whole study sample (n=175), that is, on data of male and female laypersons. In contrast, the calculations of Study 2 only rely on answers of female laypersons (n=105).

Procedure

After the local research ethical committee had accepted our project, a letter was sent to all randomly selected adult men and women to inform them about the study. Adolescent men and women of between 16 and 20 years were informed by a psychologist who came to their school. In addition, they received a letter each to read at home. Interested adolescent and adult individuals were asked to return a signed consent form. After a few months, a reminder was sent to everyone that had not yet answered.

Whereas adults came to the hospital for the survey, participants of the adolescent group were interviewed at their school in small groups of 5-10 persons. Young men and women who were below the legal age of majority at the time of the study (younger than 18 years old) had to have a consent form signed by their parents to participate.

After answering the standardized questionnaire, participants had the opportunity to talk to a psychologist.

Measures

We developed a standardized questionnaire which assessed a number of topics. First, laypersons were asked questions regarding the importance of several aspects of penile appearance in general. Second, they were asked about socio-economic data, their own sexuality and their genital self-perception in order to identify perceiver-related characteristics that are relevant predictors of their perception of HASRGs. Third, 10 standardized photographs of non-erect HASRGs (six with a distal type of hypospadias and four with a penile or a more proximal form of hypospadias) and 10 photographs of non-erect circumcised penises (controls) were presented to laypersons to measure how they rated these genitals. Circumcised men were chosen as controls because they have a similar penile appearance (due to the absent foreskin) as men with a corrected hypospadias. Every photoset contained four photos with standard views of the non-erect penis (see Figure 6). They were made by a medical photographer at the University Children's Hospital. Rating laypersons were informed that they had to evaluate photos of surgically repaired genital malformations. They did not know that half the photosets showed "normal", circumcised genitals. The development of the photosets and more details of the photographed men with corrected hypospadias or with circumcised penis are described in sections B1 & B2.

In the self-developed questionnaire, data were assessed by the following measures.

All the measures described below and all statistical analyses applied are described in more detail in sections B1 & B2. Figure 5 illustrates the variables and outcome measures assessed in study A.

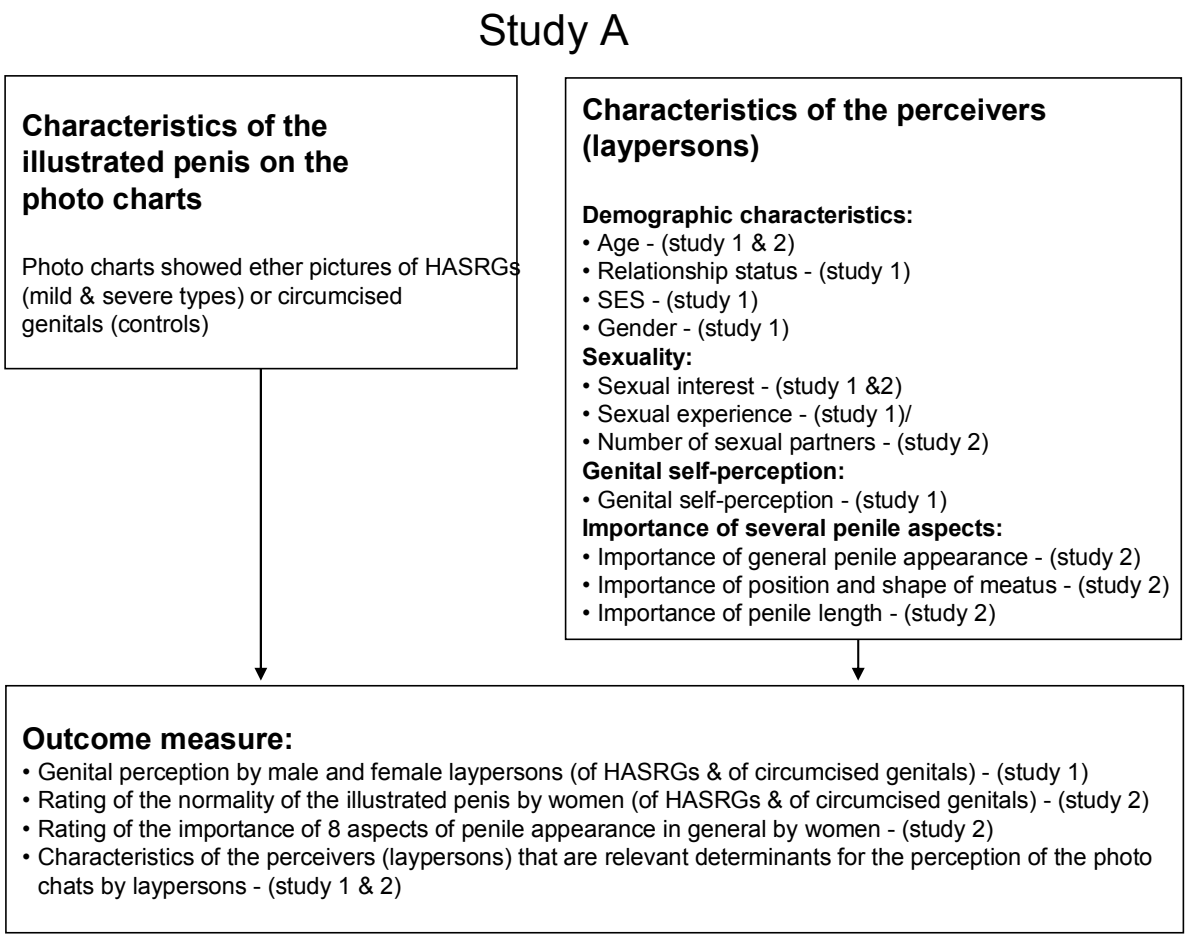

Figure 5: Schematic diagram of the variables and outcome measures assessed in Study A

Measures to assess how laypersons perceive HASRGs compared to non-affected, circumcised genitals

Figure 6: Example of a photoset of a man with corrected distal hypospadias which was presented to the rating laypersons.

Reprinted from Weber et al. (2013) with permission from Elsevier

- Self-developed question to assess the evaluation of each photoset: Normality of penile appearance (see question 1 below)

Question 1: To assess the rating of the normality of the penile appearance

How much do you agree with the following statement?

This is a normal (-looking) penis

- ❑ I fully agree
- ❑ I rather agree
- ❑ I rather disagree
- ❑ I fully disagree

- “Mean score of laypersons’ genital perception” to assess the evaluation of each photoset: Evaluation of satisfaction with aspects of the photographed penis (the mean score is based on the PPS by Weber et al. (2013) or on the work of Mureau, Slijper, Slob, et al. (1995) and Winter (1989) - see question 2 below)

Question 2: To calculate the mean score of laypersons’ genital perception

How satisfactory are the following penile aspects of the illustrated penis?

	Very satisfactory	Satisfactory	**Un-**satisfactory	Very **un-**satisfactory
a) Penile length	❑ 4	❑ 3	❑ 2	❑ 1
b) Penile girth	❑ 4	❑ 3	❑ 2	❑ 1
c) Position and shape of meatus	❑ 4	❑ 3	❑ 2	❑ 1
d) Shape of glans	❑ 4	❑ 3	❑ 2	❑ 1
e) Appearance of scrotum	❑ 4	❑ 3	❑ 2	❑ 1
f) Shape of penile skin	❑ 4	❑ 3	❑ 2	❑ 1
g) Appearance of pubic hair	❑ 4	❑ 3	❑ 2	❑ 1

Measures to assess perceiver-related characteristics which are predictors for laypersons’ genital perception of HASRGs

- Sexual Interest Scale (The Sexual Interest Scale was developed on the basis of the work of Bucher, Hornung, Gutzwiler, and Buddenberg (2001))

- Sexual Experience Scale (developed for this study)

- Question on number of sexual partners (developed for this study)

- Question on genital self-perception (developed for this study and based on the PPS (Weber et al., 2013))

- Question on socio-economic status (SES) (Socioeconomic scale by Largo, Molinari, Comenale Pinto, Weber, and Duc (1986))

Measure to assess which aspects of penile appearance are considered to be relevant

- Question rating the importance of different aspects of penile appearance (developed for this study and based on the Penile Perception Score (PPS) (Weber et al., 2013) or on the work of Mureau, Slijper, Slob, et al. (1995) and Winter (1989) – see question 3 below)

Question 3: To assess the rating of the importance of several penile aspects

How important do you consider the following aspects of a penis in general?

	very important	important	neither important nor un-important	**un-** important	very **un-** important
a) penile length	❑ 5	❑ 4	❑ 3	❑ 2	❑ 1
b) penile girth	❑ 5	❑ 4	❑ 3	❑ 2	❑ 1
c) position and shape of meatus	❑ 5	❑ 4	❑ 3	❑ 2	❑ 1
d) appearance of glans	❑ 5	❑ 4	❑ 3	❑ 2	❑ 1
e) appearance of scrotum	❑ 5	❑ 4	❑ 3	❑ 2	❑ 1
f) appearance of penile skin	❑ 5	❑ 4	❑ 3	❑ 2	❑ 1
g) appearance of pubic hair	❑ 5	❑ 4	❑ 3	❑ 2	❑ 1
h) general cosmetic appearance	❑ 5	❑ 4	❑ 3	❑ 2	❑ 1

5.2. Research project B: Health-related quality of life in men living with corrected hypospadias

5.2.1. Questions and hypotheses

As mentioned above, long-term psychological outcome studies of hypospadias repair (especially on HRQoL) are important because scientific evidence of the positive effect of early surgery on psychological development is scarce.

Only two previous studies regarding the HRQoL of adults with corrected hypospadias used validated measures. However, they did not compare their results with those from a control group. Therefore, the aim of the present study was to investigate the HRQoL of adults with corrected hypospadias by means of a validated questionnaire in comparison to a control group. In addition, we wanted to find the most relevant demographic, medical and sexual variables which predict a better HRQoL. Thus, the following research questions were developed.

Research Question 3a:

Do men with a corrected hypospadias have impaired HRQoL in comparison to non-affected, circumcised controls?

Research Question 3b:

Which are the most relevant determinants of hypospadias patients' HRQoL?

Studies have shown that men with corrected hypospadias more often report erectile dysfunction, ejaculation difficulties and a more negative genital self-perception than non-affected controls (Rynja, de Jong, et al., 2011).

Research Hypothesis 3a:

Therefore, we hypothesized that men with a corrected hypospadias have impaired HRQoL.

Hypothesis 3b is based on the same findings as hypothesis 3a and on the assumption that being in an intimate relationship, which offers some kind of social support, and a mild type of hypospadias, which possibly causes less stress than a severe type of hypospadias leads to higher HRQoL. Furthermore, it was created in light of a study in children and adolescents which showed that higher age was associated with a better HRQoL.

Research Hypothesis 3b:

Therefore, the following variables were expected to be predictors of a better HRQoL: being in an intimate relationship, increasing age, a mild type of hypospadias, a more favorable genital self-perception, and a better orgasmic and erectile function.

5.2.2. Methods

Study design

This cross-sectional study investigated the HRQoL of men with corrected hypospadias in comparison with that of circumcised men (controls). Particular attention was paid to variables which predict a better HRQoL of men with corrected hypospadias.

Subjects

Subjects were recruited in two ways. In a first step, every volunteer who let his penis be photographed for the development of the questionnaire for Study A (see chapter A 5.1.) also received questionnaires (for Study B) concerning their HRQoL, their sexuality, their genital self-perception, and their socio-demographic data to complete at home. A total of 16 of 20 men with corrected hypospadias and 16 of 18 circumcised men who let their penis be photographed also completed these questionnaires. In a second step, questionnaires for Study B were sent to every man that did not participate as a volunteer for the photographs in Study A. Thus, questionnaires were sent to 198 men with corrected hypospadias and 310 circumcised men. Circumcised men were chosen as controls because their penile appearance is similar to that of men with corrected hypospadias and therefore allow a measurement of the pure effect of hypospadias.

In both steps of the recruitment, 11 hypospadias patients and 21 circumcised men could not be contacted. In total, 52 (25.1%) hypospadias patients and 46 (15.0 %) circumcised men completed the SF-36 questionnaire. All participants were older than 18 years and had been

treated in their childhood at the University Children's Hospital, with the exception of one hypospadias patient and three circumcised men who had been operated elsewhere. Seven hypospadias patients who completed the SF-36 questionnaire were excluded for one of the following exclusion criteria: a chronic disease or disability, persistent uncorrected hypospadias or other findings for an intersex disorder besides hypospadias.

Procedure

After approval by the local research ethical committee, all participants of Study A received questionnaires to fill out at home. In a next step, questionnaires and an information-letter about the HRQoL- study were sent to patients and controls that did not take part in Study A. Interested men were asked to return the completed questionnaires and a signed consent form.

Measures

To evaluate hypospadias patients' HRQoL and potential predictors of a better HRQoL, the following standardized measures were applied. All measures and statistical analyses applied are described in more detail in section B3. Figure 7 illustrates the study design.

Method to measure health-related quality of life (HRQoL)

- The Medical Outcomes Study Short Form-36 item questionnaire (SF-36) (Ware et al., 2000)

Measures to assess potential predictors of a better HRQoL of men with corrected hypospadias

- The Penile Perception Score (PPS) (Weber et al., 2013) to measure hypospadias patients' genital self-perception

- Erectile function (EF) (Subscale of the International Index of Erectile Function (IIEF) (Rosen et al., 1997))

- Orgasmic function (OF) (Subscale of the International Index of Erectile Function (IIEF) (Rosen et al., 1997))

- Socio-economic status (SES) (Socioeconomic scale by Largo et al. (1986))

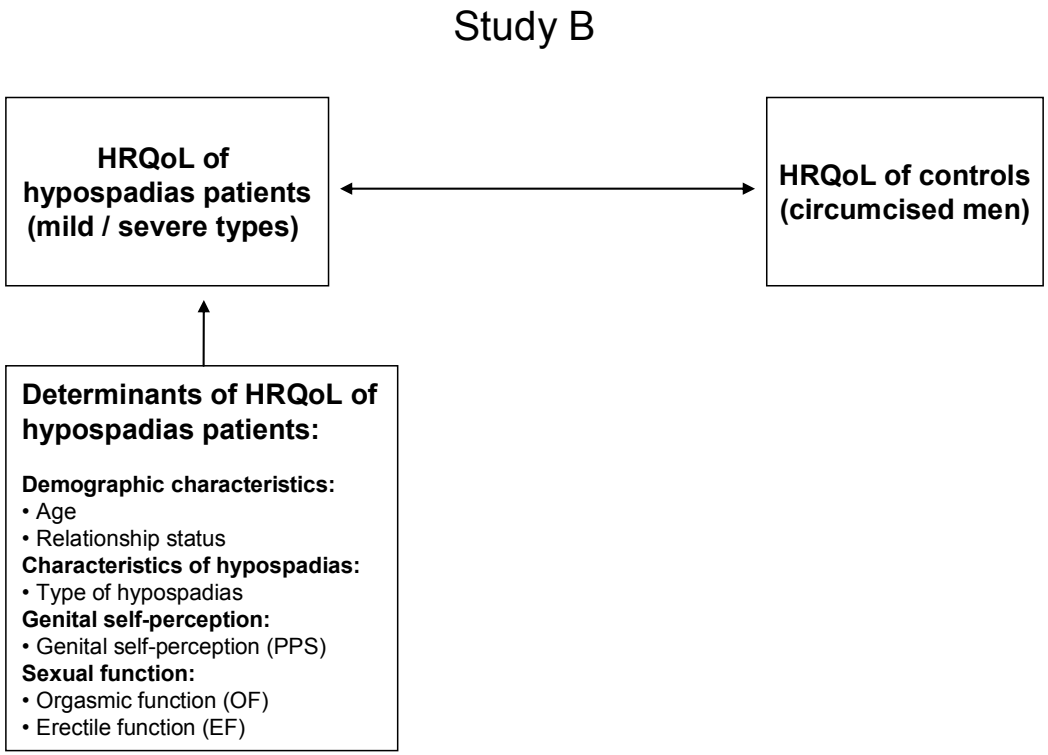

Figure 7: Schematic diagram of variables and outcome measures assessed in study B

5.3. Outline of the empirical research section

The present doctoral thesis contains three empirical papers (B1, B2, B3). Section B1 presents findings of a cross-sectional study on how laypersons unacquainted with hypospadias evaluate HASRGs in comparison with non-affected, circumcised penis. Furthermore, it identifies laypersons' characteristics that predict a better evaluation of HASRGs.

> Ruppen-Greeff, N.K., Landolt, M.A., Gobet, R. & Weber, D.M. (to be submitted). Appraisal of adult genitalia after hypospadias repair: Do laypersons mind the difference?

Section B2 describes the results of a comparative study investigating whether women consider HASRGs as normal-looking compared to non-affected, circumcised genitals. Again, perceiver-related characteristics are identified, which determine whether HASRGs or a circumcised penis is considered as normal-looking or not. Moreover, the study investigates how relevant women consider single aspects of penile appearance.

> Ruppen-Greeff, N.K., Weber, D.M., Gobet, R., Landolt, M.A. (2015). What is a good looking penis? How women rate the penile appearance of men with surgically corrected hypospadias. *Journal of Sexual Medicine, 12*(8), 1737-1745.

Section B3 presents the results on HRQoL of men with corrected hypospadias in comparison with circumcised men. Furthermore, determinants of hypospadias patients HRQoL were examined.

> Ruppen-Greeff, N.K., Weber, D.M., Gobet, R. & Landolt, M.A. (2013). Health-related quality of life in men with corrected hypospadias: An explorative study. *Journal of Pediatric Urology, 9*(5), 551-8.

Finally, section C contains a summary and overall discussion of the results and suggestions for future research activities and clinical practice.

Part B Empirical Research Section

1. Appraisal of adult genitalia after hypospadias repair: Do laypersons mind the difference?

1.1. Abstract

Introduction

Men with corrected hypospadias often suffer from sexual inhibition and fear of being ridiculed by others because of their penile appearance. However, no investigation has thus far been made of the perception of hypospadias-affected surgically-repaired genitals by laypersons unacquainted with hypospadias. Therefore, the aim of this study was to find out whether laypersons notice a difference between genitals of men with corrected hypospadias in comparison with circumcised genitals. Furthermore, the most relevant predictors of laypersons' perception of hypospadias-affected genitals were examined.

Study design

A cross-sectional study was performed in which a questionnaire with 10 standardized photographs of non-erect hypospadias-affected genitals and 10 circumcised genitals was presented to laypersons unacquainted with hypospadias to measure how they rated these genitals. Laypersons were 105 women and 70 men of three different age groups (age ranges 16-20, 25-30 and 40-45 years). Furthermore, laypersons were asked about demographic characteristics, their sexuality and their genital self-perception.

Results

Results showed that genitals with distal forms of hypospadias were rated similarly to circumcised genitals. In contrast, genitals with more proximal types were perceived as significantly less positive than circumcised genitals. However, the effect size was small. Higher age, being in an intimate relationship, higher socio-economic status and a higher sexual interest predicted a better layperson's perception of hypospadias-affected genitals.

Discussion

These findings do not support the fear of some men with corrected hypospadias of being ridiculed by others because of their penile appearance. Results indicate that laypersons do not notice a difference between corrected distal types of hypospadias (which represent the majority of hypospadias) and circumcised genitals. Although the findings showed that

laypersons perceive more proximal forms of hypospadias less positive than circumcised genitals, the difference does not appear to be clinically relevant as the effect size was small.

A major strength of this study is its comprehensive study design. However, the low response rate of hypospadias patients and controls for photo documentation and of laypersons who rated these photosets is a limitation of the study. Therefore, generalization from the results must be made carefully.

Conclusions

Results are relevant for patient counseling. Knowing that the penile appearance would not trouble laypersons may prevent the development of a negative genital self-perception and feelings of shame. The findings also suggest that hypospadias-affected genitals seem to be rated more positively when laypersons know more about the "normal" variation of penile appearance (e.g. with increasing sexual experience at a higher age).

1.2. Introduction

Although much research on hypospadias has been published over the last 10 years, few studies have addressed the perception of hypospadias-affected surgically-repaired genitals by the patients themselves or by others.

However, these studies have clearly shown that men with corrected hypospadias (particularly those with a proximal hypospadias) are more dissatisfied with their genital appearance than control subjects (Mureau, Slijper, van der Meulen, et al., 1995; Ortqvist et al., 2014; Rynja, de Jong, et al., 2011; Rynja et al., 2012; Wang et al., 2010). In addition, men with corrected hypospadias are more often inhibited in seeking sexual contact (Mondaini, Ponchietti, Bonafè, et al., 2002; Mureau, Slijper, van der Meulen, et al., 1995; Wang et al., 2010) or are afraid of being ridiculed by others because of their penile appearance (Wang et al., 2010).

As modern hypospadias repair aims at an anatomical reconstruction and usually achieves a good cosmetic result, it is likely that laypersons unacquainted with hypospadias do not notice the malformation after repair and that patients' inhibitions due to their penile appearance may be unfounded. Thus far, no investigation has assessed whether laypersons actually notice the malformation after hypospadias repair. The perception of hypospadias-affected genitals has been investigated only among medical personnel, parents and intimately involved persons (e.g. Mureau et al., 1996; Weber et al., 2008).

Therefore, our study focused on how laypersons perceive hypospadias-affected surgically-repaired genitals in comparison to non-affected, circumcised genitals. Furthermore, the most relevant predictors of laypersons' perception of hypospadias-affected genitals were identified.

1.3. Material and methods

A cross-sectional study was performed in which a questionnaire with standardized photographs of non-erect hypospadias-affected surgically-repaired genitals and circumcised genitals was presented to laypersons unacquainted with hypospadias to measure how they rated these genitals.

1.3.1. Subjects – laypersons asked to rate hypospadias-affected genitals

Subjects were women and men without previous knowledge about hypospadias who had no affiliation to the hospital or to any of the patients. They were recruited to three different age groups (age ranges 16-20, 25-30 and 40-45 years); it was assumed that these correlate with different degrees of sexual experience.

The youngest age group (16-20 years) was recruited at schools of two different educational levels. Twenty-five of 305 young men (8.20%) and 33 of 136 young women (24.26%) agreed to participate.

Furthermore, two randomly selected population samples of men and women (25-30 years or 40-45 years old) were ordered from a local database agency. Of 800 women and 800 men contacted, 25 women (3.13%) and 38 men (4.75%) were untraceable. Of the remainder, 41 men (5.38%) and 66 women (8.25%), as well as 6 women and 4 men who were partners or friends of those invited, agreed to participate.

1.3.2. Measures

Development of photosets for laypersons

Men with hypospadias-affected surgically-repaired genitals and men with circumcised genitals who were operated in their childhood were asked permission to take photographs of their genitals. An information letter about the study was sent with a consent form to a random sample of 218 men with treated hypospadias with a present age older than 18 years who had been treated for hypospadias. Nineteen out of these 218 (8.72%) men with treated

hypospadias agreed to participate. Circumcised men were chosen as controls because they have a similar penile appearance to men with corrected hypospadias. A randomly selected group of circumcised men (n=328) who were older than 18 years were eligible as controls. Twelve of these circumcised men (3.66%) and 6 other circumcised men agreed to participate. Photosets with 4 standard views of the non-erect penis were made by a medical photographer (Figure 8).

A standardized questionnaire was developed to assess laypersons' perception of those photosets. A number of twenty photosets were selected in order to keep the survey in a tolerable length. Ten of these photosets showed circumcised genitals and were each matched with a photo of a hypospadias-affected genital. Photosets were paired according to the attributes of penile size, age, and body weight. Scars and configuration of the meatus (all were glanular) were considered as representative to the cohort by urologists among the authors. Because our pictures showed non-erect genitals, persistent curvatures were not visible. The survey was piloted to ensure that there was no confusion with the terminology of the questionnaire and to see whether the survey was feasible.

Table 2 shows the demographic and medical characteristics of the hypospadias patients and the circumcised controls selected for photosets.

Figure 8: Photo chart as presented to women (this example illustrates distal hypospadias)

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Table 2: Demographic and medical characteristics of hypospadias patients and circumcised controls selected for photosets

	Men with corrected hypospadias (n=10)		**Men with circumcised genitals (controls) (n=10)**	
Age at participation				
Mean (SD)	24.00 y	(2.45)	30.40 y	(7.34)
Median (range)	24.00 y	20-28 y	30.50 y	22-44 y
Type of hypospadias				
Distal	6			
Penile and Proximal	4			
Age at first operation				
Mean (SD)	2.67 y	(1.98)	5.56 y°	(4.75)°
Median (range)	2.79 y	0.00-6.83	2.17 y°	0.83-10.83°
Age at last operation *				
Mean (SD)	4.07 y	(2.70)		
Median (range)	3.13 y	1.08-10.33		
Surgical technique				
MAGPI technique	3			
Mathieu technique	3			
Island flap technique	2			
Onlay flap technique	1			
Denis Brown technique	1			
Number of operations				
One procedure	7		10	
Two procedures	3		0	

* Hypospadias patients were operated between 1980 and 1994.

° Valid n = 7

Laypersons' perception of photosets

Laypersons were informed that every photoset showed a surgically repaired penile malformation. In fact, every questionnaire included ten photosets of non-erect hypospadias-affected genitals and ten photosets of circumcised, "normal" genitals in a random order. One half of the participants (n=91) received photosets in reversed order.

The term "laypersons' genital perception" describes laypersons' satisfaction with different

aspects of a photographed genital. Laypersons' genital perception of each photoset was rated with seven items that evaluate satisfaction with different aspects of a penis on a four-point scale: very unsatisfactory (1), unsatisfactory (2), satisfactory (3), very satisfactory (4). Items were: penile length, penile girth, position and shape of meatus, shape of glans, appearance of scrotum, shape of penile skin, and appearance of pubic hair. They are based either on the Penile Perception Score (PPS) (Weber et al., 2013) or on the work of Mureau, Slijper, Slob, et al. (1995) and Winter (1989). In a first step, for every genital perception item (e.g. penile length, shape of glans) a mean score was calculated for photosets of hypospadias-affected genitals and for photosets of circumcised genitals. In a second step, a mean score across the seven genital perception items was calculated for each condition (hypospadias and circumcision). These variables were labelled: "Mean score of laypersons' genital perception of hypospadias-affected genitals" and "Mean score of laypersons' genital perception of circumcised genitals". The internal consistency of both mean scores of laypersons' genital perception was good (Cronbach's Alpha > .85).

Self-reported genital perception of laypersons

Besides rating photosets, participants were asked to assess their own genitals. They had to express satisfaction with their own genital appearance on a 4-point scale: very dissatisfied (1), dissatisfied (2), satisfied (3), very satisfied (4).

Sexuality of laypersons

Furthermore, participants were asked about their own sexuality.

On the basis of the work of Bucher et al. (2001) we developed a short Sexual Interest Scale that was calculated by the averaged sum of two items. First, participants had to express their level of sexual desire to e.g. have sexual intercourse during the last 3 months. Second, they were asked how often they had masturbated during the last 3 months. Both items were scored as follows: 0= never, 1= less than once a month, 2= once a month, 3= twice or 3 times a month, 4= once a week, 5= twice or 3 times a week, and 6= every or nearly every day.

Participants were also asked if they had any experience in petting and sexual intercourse. Both questions could be answered with yes (=1) or no (=0). The sum of the scores of both questions comprised the Sexual Experience Scale.

Socio-economic status (SES)

SES for adults was calculated on the basis of their education and occupation, and SES for adolescents was calculated on the basis of maternal education and paternal occupation. Education and occupation were scored on a 6-point scale. SES scores (range: 2-12 points) were subdivided into three social classes: lower class (2-5 points); middle class (6-9 points), and upper class (10-12 points). This measure has proven to be a reliable and valid indicator of SES (Landolt, Vollrath, Gnehm, & Sennhauser, 2009).

1.3.3. Procedures

After approval of the study by the local ethical committee, an information letter about the study was sent to all the laypersons selected, and they were asked to return a signed consent form. After a few months a reminder was sent to all non-responders.

Adolescents were interviewed at school and adults at the hospital.

1.3.4. Statistical analysis

Data were analyzed with SPSS (Version 16) for Windows (SPSS Inc., Chicago, IL). T-tests were used to compare groups on continuous data. Because most variables were ordinal or categorical, nonparametric statistics were applied as Spearman-Brown rank correlations, Mann-Whitney *U* tests, Wilcoxon tests or χ^2 -tests with $p< 0.05$ considered significant. Effect sizes (Cohen's d) were calculated to show clinical relevance of the group differences: 0.20= small effect sizes; 0.50= medium effect sizes; >0.80 large effect sizes. A multiple regression analysis was computed with a normally distributed dependent variable according to the Kolmogorov-Smirnov test. The dependent variable was "Mean score of laypersons' genital perception of hypospadias-affected genitals". The predictors were chosen according to their significance in bivariate correlations with the dependent variable and based on a priori assumptions. They were entered blockwise into the regression analyses. Laypersons' demographic data was entered as block 1: male sex, age, living in an intimate relationship and SES. Laypersons' sexuality was entered as block 2: sexual interest and sexual experience. Laypersons' genital self-perception was entered as block 3.

1.4. Results

1.4.1. Descriptive statistics of the sample

Table 3 shows the demographic and sexual characteristics of the final study sample (n=175), consisting of 105 women and 70 men who answered the questionnaire on genital perception. No significant differences were observed in men's and women's genital self-perception, their demographic characteristics or their degree of sexual experience. However, men had a significantly higher sexual interest than women.

1.4.2. Laypersons' rating of the photos

A Wilcoxon-test showed that laypersons rated the mean score of genital perception of hypospadias-affected genitals (mean: 2.65, SD=0.37) significantly (p= 0.001) inferior to that of circumcised genitals (mean: 2.71, SD=0.40). The effect size was very small (Cohen's d= -0.16). Regarding the severity of hypospadias, the mean score of genital perception of more proximal forms of hypospadias (mean: 2.58, SD=0.38) was rated as significantly less positive than that of distal types of hypospadias (mean: 2.69, SD=0.37; p= 0.000; Cohen's d= -0.29) and of circumcised genitals (p= 0.000; Cohen's d= -0.33). However, effect sizes were small. No significant difference in the mean score of laypersons' genital perception could be found between distal forms of hypospadias and circumcised genitals (p= 0.99). The effect size was very small (Cohen's d = -.05).

1.4.3. Factors relating to laypersons' rating of the photos of hypospadias-affected genitals

Table 4 shows the multiple regression analysis that evaluated the relationship between laypersons' perception of hypospadias-affected genitals and several other variables of the laypersons. The regression model for predicting laypersons' genital perception of hypospadias-affected genitals was significant. The predictors explained 10% of the variance of the dependent variable. There were four significant predictors of a better laypersons' perception of hypospadias-affected genitals: higher age, being in an intimate relationship, higher socio-economic status (SES) and a higher sexual interest. In other words, a layperson who had these attributes was more likely to rate hypospadias-affected genitals positively.

Table 3: Demographic characteristics and sexual characteristics of the laypersons sample (n =175)

	Women (n=105)		Men (n=70)		p†
Age					
Age category (16-20y)	33	31.4%	25	35.7%	0.84
Age category (25-30y)	44	41.9%	22	31.4%	
Age category (40-45y)	28	26.7%	23	32.9%	
Mean (SD)	28.37y	9.65	28.67y	10.17	
Median (range)	27.00	16-45	27.50	16-45	
Socio-economic status (n)*					
Lower	7	6.6%	3	4.3%	0.93
Middle	51	48.6%	38	54.3%	
Upper	47	44.8%	29	41.4%	
Relationship status (n)					
Living in an intimate relationship	65	61.9%	46	65.7%	0.61
Single	40	38.1%	24	34.3%	
Sexual Interest Scale					
Mean (SD)	3.44	1.29	4.98	0.98	**0.000**
Median (range)	3.50	0.0-6.0	5.50	2.5-6.0	
Sexual Experience Scale					
Experience in petting					
No	12	11.4%	11	15.7%	0.41
Yes	93	88.6%	59	84.3%	
Experience in sexual intercourse					
No	15	14.3%	12	17.1%	0.61
Yes	90	85.7%	58	82.9%	
Genital self-perception					
Mean (SD)	3.17	0.51	3.32	0.56	0.06
Median (range)	3	2-4	3	2-4	

† T-tests for continuous data; U test according to Mann-Whitney for ordinal data; χ^2-tests for categorical variables.

* U test according to Mann-Whitney on the basis of SES sum scores.

Table 4: Regression analysis on the mean score of laypersons' genital perception of hypospadias-affected surgically-repaired genitals

Variable	B	Standard Error of B (SEB)	Beta	R^2	Difference in R^2
Block 1:					
Laypersons' demographic data				0.11**	
Male Sex	-0.05	0.07	-0.06		
Age	0.01	0.00	0.20*		
Living in an intimate relationship	0.17	0.07	0.23*		
SES	0.03	0.01	0.18*		
Block 2:					
Laypersons' sexuality				0.14**	0.03
Sexual Interest	0.05	0.02	0.19*		
Sexual Experience	-0.07	0.05	-0.12		
Block 3					
Laypersons' genital self-perception				0.14**	0.00
Self-perception	0.03	0.05	0.05		

Equation: R = 0.37 (n = 165, F = 3.61, p = 0.001), R^2 = 0.14, R^2 adjusted = 0.10; *$p<0.05$, **$p<0.01$

1.5. Discussion

The current study indicates that the fear of some hypospadias patients of being ridiculed because of their penile appearance (Wang et al., 2010) is not justified. We found that laypersons unacquainted with hypospadias rated genitals of men with distal forms of hypospadias (which represents the majority of hypospadias) similarly to the genitals of non-affected, circumcised men. Although the less common and more proximal forms of hypospadias were not perceived as positively as circumcised genitals, the difference is most probably not clinically relevant. Notably, the effect size was small. In other words, it seems that the penile appearance of corrected proximal hypospadias does not trouble laypersons much.

The result that the more proximal forms of hypospadias were rated less positively than the distal types of hypospadias seems to meet patients' expectations. Previous studies have shown that a lower genital self-perception was associated with more proximal hypospadias (Chertin et al., 2013; Liu et al., 2006; Mureau, Slijper, van der Meulen, et al., 1995; Ortqvist et al., 2014; Rynja, de Jong, et al., 2011; Rynja et al., 2012). Furthermore, men with proximal hypospadias reported hearing silly remarks or being ridiculed because of their penile appearance more often than those with distal forms (Liu et al., 2006).

In contrast to our results, Mureau, Slijper, Nijman, et al. (1995) reported that more hypospadias patients than controls had received comments regarding the appearance of their genitals. However, it is not clear whether it is the penile appearance or rather the inhibited behavior of the patients (e.g. in public toilets) that may have led to those remarks. In line with our results, a recently published study (Vandendriessche et al., 2010) found that adolescents with operated hypospadias had not been ridiculed significantly more often than non-affected controls.

The present study also examined predictors of a positive laypersons' genital perception of hypospadias-affected genitals. We found that higher age, being in an intimate relationship, higher socio-economic status, and a higher sexual interest of the raters predicted a better laypersons' genital perception of hypospadias-affected genitals. Possibly, laypersons develop a more realistic body image with increasing sexual experience at a higher age, when being in an intimate relationship and when being sexually interested, as a consequence of the confrontation with the imperfections of a partner's body. A positive association between better genital perception and increasing age (Bramwell & Morland, 2009) and a correlation between a positive genital perception and higher sexual interest (respectively sexual desire) was also found in studies on the genital self-perception of women (Ålgars et al., 2011).

Limitations

A major strength of the current study is its comprehensive design. However, the low response rate is a limitation of the study. Therefore, it is likely that the selected photosets do not represent the whole variation in outcomes of hypospadias repair. However, the selected photosets do represent a wide spectrum of different types of hypospadias. In addition, hypospadias patients and circumcised controls who accepted photo documentation of their genitals possibly had a better surgical result than the non-responders, resulting in a selection bias. Yet, the urologists among the authors did not consider the results in photosets to be particularly favorable. On the contrary, they assume that patients treated with current techniques have better long-term outcomes than those in the study, who were operated up to 32 years ago. Furthermore, laypersons who are open minded about sexuality may be overrepresented (particularly with the low response rate of laypersons in our study). In addition, middle and upper socio-economic status were overrepresented. Therefore, generalization from the results must be made carefully. Pictures of hypospadias patients were compared with those of circumcised men. Therefore, the current results do not allow generalization to non-circumcised men. Finally, the "mean score of laypersons' genital perception" was not validated.

Clinical implications

The findings of this study are relevant for patient counseling. Specifically, the knowledge that the appearance of hypospadias-affected genitals does not trouble laypersons should be communicated to the patients and their parents to prevent the development of shame, especially because feelings of shame about penile appearance (Schonbucher et al., 2008a) and a negative genital perception (Ruppen-Greeff, Weber, Gobet, & Landolt, 2013) are risk factors for an impaired health-related quality of life (HRQoL) of hypospadias patients. Therefore, we recommend a comprehensive brief counseling about hypospadias not only for the patients, if old enough, but also for the parents. This is important because the emotional reaction of the parents towards the hypospadias seems to be of crucial importance for the psychological development of the young patients (Easson, 1966). Furthermore, we recommend a late follow-up in adolescence or even beyond puberty, as has been suggested by several authors (e.g. Bracka, 1989; Ekmark, Svensson, Arnbjornsson, & Hansson, 2013; Schonbucher et al., 2008b; Springer, 2014). This ensures that patients, who are usually operated at an age when they are not able to understand counseling, receive this information from a professional and have the opportunity to ask any question or address concerns regarding their genitals. Various studies have indicated the importance of comprehensive information (e.g. Mondaini, Ponchietti, Bonafè, et al., 2002; Schonbucher et al., 2008b) and

of a late follow-up (e.g. Aho et al., 1997; Bracka, 1989; Ekmark et al., 2013) for a good adaptation to the penile condition.

Interestingly, studies (Dodds et al., 2008; Fichtner et al., 1995) of men with non-operated hypospadias claim that many of them, some of their sexual partners (Fichtner et al., 1995), and even some professionals (Donaruma-Kwoh, Tran, & Giardino, 2010) were not aware of the hypospadias. Therefore, it would be interesting to investigate how laypersons perceive non-operated genitals. It is quite possible that they too are not troubled by the appearance of non-operated distal hypospadias. In line with Fichtner et al. (1995) and Dodds et al. (2008), this would raise the fundamental question whether aesthetic correction of distal hypospadias is indicated in early childhood or whether routine surgical correction should be limited to severe forms that impair sexual function.

Conclusions

In conclusion, the findings of the current study do not support hypospadias patients' fear of being ridiculed or rejected by others because of their penile appearance. Corrected distal forms of hypospadias (which represent the majority of hypospadias) received a similar rating by laypersons to those of circumcised genitals. More proximal forms of hypospadias had a more negative genital perception by laypersons than circumcised genitals. However, the difference does not appear to be clinically relevant as the effect size was small.

In addition, results suggest that laypersons' perception of penile appearance may vary due to several influences. Notably, we found that hypospadias-affected genitals seem to be rated more positively when laypersons know more about the "normal" variation of penile appearance (e.g. with increasing age).

As a consequence for clinical management, we suggest that hypospadias patients and their parents learn about the results of this study. The knowledge that the penile appearance would not trouble laypersons may help prevent the development of a negative genital self-perception and feelings of shame.

2. What is a good looking penis? How women rate the penile appearance of men with surgically corrected hypospadias.

2.1. Abstract

Introduction

Some men with corrected hypospadias perceive their penile appearance to be abnormal, although health professionals consider these results satisfactory.

The aim of this study was to investigate how relevant women consider single aspects of penile appearance to be. Moreover, we studied whether women perceive hypospadias-affected surgically-repaired genitals (HASRGs) to be as normal-looking as circumcised genitals and identified the most relevant predictors that influence whether a penis is perceived as normal.

Methods

In this cross-sectional study, 105 women in different age groups (age range: 16-20; 25-30 and 40-45 years) completed a standardized questionnaire.

Participants had to rate the importance of eight penile aspects and to indicate how normal they found the appearance of standardized photos of 10 HASRGs and of 10 circumcised genitals. Furthermore, they were asked about demographic characteristics and their sexuality.

Results

Results showed that women considered the position and shape of the meatus as the least important penile aspect. Furthermore, results showed that HASRGs with distal hypospadias were perceived to be as normal-looking as circumcised genitals, whereas genitals with more proximal hypospadias were perceived as significantly less normal. However, the difference need not be considered clinically relevant, as the effect size was small. Observer-related predictors of a more positive penile perception were higher age, higher sexual interest, and perceiving the general cosmetic penile appearance as more important and penile length as less important.

Conclusions

Overall, women were found to consider the “position and shape of the meatus” as the least important penile aspect. These findings may stimulate reflections regarding the relevance of surgical correction of the meatus in minor forms of hypospadias.

In addition, this study indicates that women perceived genitals of men with distal operated hypospadias (which represents the majority of hypospadias) to be as normal as non-affected, circumcised genitals.

2.2. Introduction

Hypospadias is one of the most common penile malformations, occurring in approximately 1 of 200 to 1 of 300 live male births (Baskin & Ebbers, 2006; Manzoni et al., 2004). It is a misplacement of the urethral meatus, which may be located at the ventral surface of the penis to the perineum. In most types of hypospadias, the prepuce is split ventrally. More severe forms of hypospadias are associated with a curvature of the penis (Baskin & Ebbers, 2006). Today in Europe and the USA, almost all types of hypospadias are routinely corrected during the first year of life (Manzoni et al., 2004) to improve micturition and sexual function and to achieve a slit-like meatus at the tip of the glans (Baskin & Ebbers, 2006; Manzoni et al., 2004).

Although satisfactory surgical results can be achieved, men with an operated hypospadias are reported to be more dissatisfied with their penile appearance (Mureau, Slijper, van der Meulen, et al., 1995; Ortqvist et al., 2014; Rynja, de Jong, et al., 2011; Rynja et al., 2012; Wang et al., 2010) and size (Ortqvist et al., 2014; Rynja, de Jong, et al., 2011) than men without hypospadias. Furthermore, some men with hypospadias-affected surgically-repaired genitals (HASRGs) are concerned about having an abnormal penile appearance (Rynja, de Jong, et al., 2011; Sommerlad, 1975). However, this fear is also common among normal young men without genital malformations (Rynja, de Jong, et al., 2011).

Up to now, it is not clear whether only hypospadias patients themselves consider the appearance of HASRGs to be less normal-looking than non-affected genitals, or whether others (e.g. potential sexual partners) do too. It is also unclear which aspects of penile appearance are considered to be relevant by women and thus play a role in perceiving HASRGs as normal.

Aims

The aim of this study was threefold: first, to investigate which aspects of penile appearance are considered to be relevant by women; second, to know whether HASRGs are perceived to be less normal-looking than circumcised genitals; and third, to identify the most relevant observer-related predictors for penile perception.

Following a study (Fichtner et al., 1995) which demonstrated that the position of the meatus in a "normal" penis varied widely, we hypothesized that women would consider the position and the shape of the meatus as less important than other penile aspects. Furthermore, we expected women to perceive HASRGs to be as normal-looking as the genitals of non-affected, circumcised men. Based on studies among non-affected men (Francken et al., 2002; Lever et al., 2006), we further believed that women would not consider penile size to be as relevant as expected by hypospadias patients themselves.

In addition, we postulated that the following variables influence women's perception of a penis positively: higher age, higher degree of sexual interest, and a higher number of sexual partners a woman has had during her life. Moreover, we expected that a women's perception of a penis is affected by the importance she gives to the attributes of a penis, such as penile size or general appearance.

2.3. Methods

This cross-sectional study was conducted as part of a larger research project on hypospadias. In the current study, women were asked to rate photosets of men with HASRGs and men with circumcised genitals. Furthermore, they had to evaluate the importance of different aspects of a penis in general.

2.3.1. Subjects

Subjects were women of three different age groups (age ranges: 16-20 years, 25-30 years, and 40-45 years), which we supposed correlate with different degrees of sexual experience. They had no affiliation to the hospital or to any of the patients.

In the youngest age group (16-20 years), 33 of 136 female pupils (24.26%) from schools of two different educational levels agreed to participate. Further, two randomly selected population samples of women (400 women aged 25-30 years, and 400 women aged 40-45 years) were ordered from a local database agency. This agency manages a large address file which may be regarded as representative of the population. Of those 800 women, 66 women (8.25%) participated in the survey, and 25 women (3.13%) were untraceable. In addition, another 6 women who were friends of the invited women participated. Finally, a total of 105 women completed the questionnaires.

2.3.2. Main outcome measures

Development of photosets for genital appraisal

Photosets with four standard views of the non-erected penises of men with corrected hypospadias and of circumcised men (controls) were made by a medical photographer at the University Children's Hospital (Figure 9). Circumcised men were chosen as controls because the absence of a foreskin means they have a similar penile appearance to men with a corrected hypospadias.

To acquire the photosets, an information letter about the study with a consent form was sent to a random sample of 218 men with corrected hypospadias and 328 circumcised men (controls). Men of both groups were older than 18 years and had been operated at the University Children's Hospital. Nineteen (8.72%) of these 218 men with corrected hypospadias, twelve (3.66%) of the 328 circumcised men, and 6 other circumcised men agreed to participate.

A standardized questionnaire included 20 photosets: 10 of circumcised genitals and 10 of HASRGs. Photosets were paired according to the attributes of penile size, age, and body weight. In the questionnaire, photosets were shown in a random order. Half of the women (n=55) received photosets in reversed order.

Six of the selected hypospadias patients had a distal hypospadias and four a penile or a more proximal form of hypospadias. They were operated between 1980 and 1994 using the following methods: MAGPI procedure (n=3), Mathieu hypospadias repair (n=3), Island Flap technique (n=2) combined with Onlay Flap technique (n=1), and Denis Brown technique (n=1). On average, the first operation was carried out when the patients were 2.67 +/- 1.98 years old (range 0.00–6.83). The final surgery was performed at the age of 4.07 +/- 2.70 years on average (range 1.08-10.33). Seven patients underwent one procedure, and three were operated twice. At the time of the current study, the patients were 20 to 28 years old, with a mean age of 24 years (SD=2.45).

In the control group, circumcised men were operated at a mean age of 5.56 years (SD=4.75; range= 0.83 -10.83; 3 missing data). Their age at participation was 30.40 +/- 7.34 years (range: 22 – 44 years).

Figure 9: Photo chart as presented to women (this example illustrates distal hypospadias)

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Genital appraisal of photosets

Women (n=105) were asked to evaluate photos of surgically repaired genital malformations. They did not know that half the photosets showed “normal”, circumcised genitals.

For every photoset in the questionnaire, women had to indicate on a 4-point scale ranging from total disagreement (1) to full agreement (4) how much they agreed with the statement: ‘This is a normal (-looking) penis’.

Importance of penile aspects

In the questionnaire, women were also asked how important they consider eight different aspects of a penis on a 5-point scale ranging from very unimportant (1) to very important (5). The penile aspects were penile length, penile girth, position and shape of meatus, shape of glans, appearance of scrotum, shape of penile skin, appearance of pubic hair, and general cosmetic appearance. These penile aspects were chosen based on the Penile Perception Score (PPS) (Weber et al., 2013) and the work of Mureau, Slijper, Slob, et al. (1995) and Winter (1989).

We were not sure whether the exposure to multiple photos of HASRGs would affect the way women looked at genitals, with a possible influence on their evaluation of particular aspects of the appearance. Consequently, one half of the women (n=55) evaluated the importance of the penile aspects before rating photosets of HASRGs and the other half (n=50) after rating photosets of HASRGs.

Sexuality of women

Furthermore, the questionnaire included questions on women's own sexuality (e.g. numbers of sexual partners). A short Sexual Interest Scale (on the basis of the work of Bucher et al. (2001)) was calculated by the averaged sum of two items, women's level of sexual desire and women's frequency of masturbation for the last 3 months. Both items were scored as follows: 0= never, 1= less than once a month, 2= once a month, 3= twice or 3 times a month, 4= once a week, 5= twice or 3 times a week, and 6= every or nearly every day.

Socio-economic status (SES)

SES for adults was calculated on the basis of their education and occupation, and SES for adolescents was calculated on the basis of maternal education and paternal occupation. Education and occupation were scored on a 6-point scale. SES scores (range: 2-12 points) were subdivided into three social classes: lower (2-5 points); middle (6-9 points), and upper (10-12 points). This measure has proven to be a reliable and valid indicator of SES (Landolt et al., 2009).

2.3.3. Procedure

The study was approved by the ethical committee of the Canton of Zurich, which is responsible for approval of all research studies. All selected women received an information letter about the study. Those who were interested were asked to return a signed consent form. Women younger than 18 years old had to provide a consent form signed by their parents to participate. After a few months, a reminder was sent to all non-responders.

Adolescents were interviewed at school and adults at the hospital. After answering the questionnaires, participants had the opportunity to talk to a psychologist.

2.3.4. Statistical analysis

Data were analyzed with SPSS (Version 16) for Windows (SPSS Inc., Chicago, IL). A $p < 0.05$ was considered significant. Two-sided tests were performed for all calculations. Effect sizes (Cohen's d) were calculated to show the clinical relevance of group differences (Cohen, 1988). Mann-Whitney U-tests were performed for comparisons of the ratings of penile aspects that were made before and after the evaluation of photosets. Wilcoxon tests were computed regarding the appraisal of photosets.

Two multiple regression analyses were computed. After square-root transformation, both dependent variables were normally distributed (Kolmogorov-Smirnov test). Dependent variables were "normality of penile look of HASRGs" and "normality of penile look of circumcised genitals". According to their significance in bivariate correlations with the dependent variable and based on a priori assumptions, the following predictors were chosen: age, number of sexual partners, sexual interest, importance of penile length, importance of position and shape of the meatus, and importance of general cosmetic appearance.

2.4. Results

2.4.1. Characteristics of the study sample

Table 5 shows the demographic and sexual characteristics of the final study sample consisting of 105 women in three different age groups with a mean age of 28.37 years (SD= 9.65). Regarding their sexual experiences, only 18 of 33 women (54.5%) in the youngest age group (16-20 years) reported that they had experience of sexual intercourse. In contrast, all participants of the two older age groups (25-30 years & 40-45 years) had experience of sexual intercourse.

Table 5: Demographic characteristics and sexual characteristics of women (n =105)

Age		
16-20 years	33	31.4%
25-30 years	44	41.9%
40-45y years	28	26.7%
Mean (SD)	28.37 y	9.65 y
Median (range)	27.00 y	16-45 y
Socio-economic status (n)		
Lower	7	6.6%
Middle	51	48.6%
Upper	47	44.8%
Relationship status (n)		
Living in a relationship	65	61.9%
Single	40	38.1%
Number of sexual partners		
Mean (SD)	7.22	7.75
Median (range)	4.5	0-40
Sexual Interest Scale		
Mean (SD)	3.44	1.29
Median (range)	3.50	0.00-6.00

2.4.2. Importance of several penile aspects

As displayed in Table 6, on average, “general cosmetic appearance” was rated as the most relevant aspect of penile appearance, “penile length” was ranked sixth, and the item “position and shape of meatus” was ranked last.

Some penile aspects were considered significantly more important by the group of women who had seen photos of HASRGs previously than women who evaluated the aspects before having seen photos of HASRGs. These items were importance of shape of glans, importance of appearance of scrotum, importance of penile skin, and importance of penile length. Effect sizes were medium. However, penile length was still ranked sixth by women that were exposed to photosets of HASRGs. Interestingly, neither the general cosmetic appearance nor

the position and shape of the meatus were perceived as more important by women before or after exposure to photosets of HASRGs.

2.4.3. Genital Appraisal of Photosets: Women's evaluation of the normality of the penile look of HASRGs

Statistical analyses showed that the genital appearance of circumcised men (M = 2.60, SD = 0.61) was perceived as significantly (p = 0.000) more normal-looking than the genital appearance of hypospadias patients (M = 2.45, SD = 0.60). Although group difference was significant, effect size was small (Cohen's d = 0.25). However, photosets of distal hypospadias (M = 2.53, SD = 0.62) were perceived to be as normal-looking as photosets of circumcised men (p = 0.16; Cohen's d = -0.11). Accordingly, individuals with more proximal hypospadias (M = 2.33, SD = 0.64) were rated less normal than those of distal hypospadias (p = 0.000; Cohen's d = -0.32) and than those of circumcised men (p = 0.000; Cohen's d = -0.43). Effect sizes were small.

2.4.4. Determinants of women's evaluation of the normality of the penile look of HASRGs

Results of the multiple regression analyses are shown in Table 7.

Both regression models were significant. The predictors explained 20% of the variance of the dependent variable "perceiving HASRGS as normal-looking" and 18% of the variance of the dependent variable "perceiving circumcised genitals as normal-looking". There were three variables that were significant predictors in both regression models: higher age of women, higher sexual interest of women, and evaluating penile length as less important.

Table 6: Rating of 8 aspects of penile appearance by women

	Rating before evaluation of photosets (n=55)			**Rating after evaluation of photosets (n=50)**				
Importance of 8 penile aspects	**Order**	**Mean**	**SD**	**Order**	**Mean**	**SD**	**P†**	**Cohens'd**
Importance of penile length	6	3.19	0.75	6	3.54	0.87	**0.02***	-0.43
Importance of penile girth	3	3.41	0.74	7	3.46	0.82	0.74	-0.06
Importance of position and shape of meatus	8	2.37	0.88	8	2.63	1.12	0.29	-0.26
Importance of shape of glans	5	3.28	0.90	3	3.71	0.90	**0.005****	-0.48
Importance of appearance of scrotum	7	3.02	1.00	4	3.69	0.85	**0.001****	-0.72
Importance of penile skin	3	3.41	1.00	2	3.89	0.89	**0.02***	-0.51
Importance of appearance of pubic hair	2	3.43	1.16	5	3.60	1.20	0.38	-0.14
Importance of general cosmetic appearance	1	3.85	0.74	1	4.06	0.73	0.11	-0.29

**p < 0.05, **p < 0.01*
† U-tests according to Mann-Whitney
Effect sizes according to Cohen: 0.20 small effect size, 0.50 medium effect size, >0.80 large effect size
1=very unimportant, 2=unimportant, 3= neither important nor unimportant, 4=important, 5=very important

Table 7: Multiple regression analyses for predictors of "normality of penile look"

Variable				
Equation 1: Normality of penile look (circumcised men)	**B**	**SEB**	**Beta**	**p**
Age	0.01	0.00	0.33	**0.001****
Number of sexual partners	0.00	0.00	0.10	0.29
Sexual interest	0.04	0.01	0.23	**0.02***
Importance of penile length	-0.06	0.03	-0.25	**0.02***
Importance of "position and shape of meatus"	-0.02	0.02	-0.12	0.22
Importance of "general cosmetic appearance"	0.05	0.03	0.19	0.11
Equation 2: Normality of penile look (hypospadias patients)				
Age	0.01	0.00	0.26	**0.009****
Number of sexual partners	0.00	0.00	0.13	0.18
Sexual interest	0.04	0.01	0.29	**0.002****
Importance of penile length	-0.07	0.03	-0.29	**0.008****
Importance of "position and shape of meatus"	-0.03	0.02	-0.15	0.12
Importance of "general cosmetic appearance"	0.06	0.03	0.24	**0.03***

*p < 0.05, **p < 0.01

Equation 1: $R = 0.48$ (n =100, F =,4.62, p **=0.000**), R^2 adj = 0.18

Equation 2: $R = 0.49$ (n =101, F =5.05, p = **0.000**), R^2 adj = 0.20

2.5. Discussion

The present research showed that there is no single penile aspect that is essential for the evaluation of penile appearance. Women considered the general cosmetic appearance as most important, while all other penile aspects were regarded as clearly less relevant.

Participating women considered the position and shape of the meatus as the least important aspect of penile appearance, although mild types of hypospadias are often operated purely to achieve a slit-like meatus at the tip of the glans. Interestingly, this did not change in the group of women who saw photos of HASRGs with less than optimal outcomes. The higher importance of the general appearance over the cosmetic aspects of the meatus has been confirmed in previous studies (Dodds et al., 2008; Fichtner et al., 1995) among men with non-operated hypospadias. Many of the affected men (Dodds et al., 2008; Fichtner et al., 1995) and their sexual partners (Fichtner et al., 1995) were not even aware of the genital malformation.

Although men with (Ortqvist et al., 2014; Rynja, de Jong, et al., 2011) and without hypospadias (Mondaini, Ponchietti, Gontero, et al., 2002) are often concerned about their penile size, in the present study, penile length was not considered as very relevant by women. Other studies have confirmed that most women find penile size “unimportant” or even “totally unimportant” (Francken et al., 2002) and that most women are satisfied with their partner’s penis size (Lever et al., 2006).

We also investigated whether women perceived HASRGs as normal-looking. Our study indicated that the genitals of men with corrected distal hypospadias (which represents the majority of hypospadias) were considered to be normal. The more proximal forms of HASRGs were evaluated as less normal-looking than circumcised genitals. However, the perceived difference is most probably not clinically relevant, as the effect size was small. In addition, more recent surgical techniques than those applied to this cohort of patients, who were operated up to 36 years ago, may lead to better surgical results.

The present study also investigated attributes of the observing women that influence their evaluation of whether a penis is to be considered as normal. In line with our expectations, the multiple regression analyses indicated that the older and the more sexually interested a

woman is, the more normal she perceives the appearance of a penis to be. Possibly, women develop a more realistic body image with increasing sexual experience at a higher age. In line with our results, a positive association between better genital perception and increasing age (Bramwell & Morland, 2009) and a correlation between a positive genital perception and higher sexual interest (or sexual desire) was also found in studies on the genital self-perception of women (Ålgars et al., 2011; Berman, Berman, & Miles, 2003).

As expected, our findings showed that the less important a woman considers penile length and the more important she considers general cosmetic appearance, the more normal she evaluates photosets of HASRGs.

Limitations

A strength of this study is its comprehensive methodological design. However, some limitations should be noted which may restrict the generalization of our results. First, due to the low response rate of men that accepted photo documentation, it is possible that participants with better surgical results were overrepresented in our photo sets. However, their surgical results were not considered to be particularly favorable by the urologists among the authors. Second, genitals of hypospadias patients were compared only with those of circumcised men and not with those of non-circumcised ones. Third, participants of middle and upper socio-economic status were overrepresented. Fourth, it is possible that mainly sexually open-minded women participated in the study. Fifth, only photosets of men with corrected hypospadias were presented to women, and no photosets of men with non - corrected hypospadias. Therefore, the meatus might well be evaluated as more important by women when the questionnaire contained such photosets of non-corrected hypospadias. Sixth, the present study only evaluated the opinions of women regarding the relevance of penile aspects and the appearance of hypospadias-affected genitals. Hence, the results may not be generalized to men.

Clinical implications

The findings of this study are relevant for patient counselling. Patients and their parents should be informed that women consider the penile appearance of distal HASRGs to be as normal-looking as that of non-affected genitals. In addition, they should know that women look rather at the overall genital appearance than at individual penile aspects such as the position and shape of the meatus or penile size. This information may help prevent the development of shame due to penile appearance and the emergence of an impaired genital

perception. Both feelings of shame about penile appearance (Schonbucher et al., 2008a) and negative genital self-perception (Ruppen-Greeff et al., 2013) were found to be risk factors for an impaired health-related quality of life (HRQoL) in hypospadias patients.

Interestingly, some case reports about non-operated hypospadias showed that the malformation was not recognized by many of the patients (Dodds et al., 2008; Fichtner et al., 1995; Katib, 2013) or their sexual partners (Fichtner et al., 1995), or even by professionals (Donaruma-Kwoh et al., 2010). Functional deficits are rare in mild forms of hypospadias (Fichtner et al., 1995), and they are often only operated to achieve a slit-like meatus at the tip of the glans. Nevertheless, hypospadias repair can lead to complications, and the treatment is assumed to be psychologically traumatic for patients (Bracka, 1999; Morgan & Mezey, 1999). Our study found that women rated the position and shape of the meatus as the least important aspect of penile appearance. In line with other authors (Dodds et al., 2008; Fichtner et al., 1995; Schlomer, Breyer, Copp, Baskin, & Disandro, 2014), the question arises whether every distal type of hypospadias benefits from routine surgical correction in early childhood. Surgical correction in early childhood could be limited to severe forms that impair sexual function, for instance. In mild forms, a simple circumcision could be considered instead of a surgical correction.

Conclusions

The present research showed that, although mild types of hypospadias are often only operated to achieve a slit-like meatus at the tip of the glans, women considered the “position and shape of the meatus” as the least important aspect of a penis. This may stimulate reflections regarding the relevance of correction of the meatus in minor forms of hypospadias, although the women participating in our study were not exposed to photos of non-corrected hypospadias.

In addition, our study indicates that most HASRGs, particularly mild distal forms, are perceived as normal genitals. As a consequence for clinical management, we suggest that patients and their parents should be informed about these results to prevent the development of shame.

3. Health-related quality of life in men with corrected hypospadias: An explorative study

3.1. Abstract

Objective:

The aim of the study was to evaluate health-related quality of life (HRQoL) of men with a corrected hypospadias in comparison to circumcised controls. Furthermore, determinants of HRQoL were examined.

Patients and methods:

In a cross-sectional study, HRQoL of 45 men with corrected hypospadias (mean-age: 26.2 years +/- 5.1) was compared with a control group of 46 circumcised men (mean-age: 25.5 years +/- 4.9). Participants answered three questionnaires: The Medical Outcomes Study Short Form-36 item questionnaire (SF-36), the International Index of Erectile Function (IIEF), and the Penile Perception Score (PPS).

Results:

Physical and mental dimensions of HRQoL were not impaired among men with corrected hypospadias if compared to circumcised men. Neither socio-demographic and medical variables nor erectile function (EF) predicted HRQoL. However, a negative genital self-perception (PPS) and a lower orgasmic function (OF) were risk factors for an impaired mental HRQoL.

Conclusions:

Our data suggest that the HRQoL of adult hypospadias patients is comparable to that of circumcised men. However, patients should be supported in developing a positive genital self-perception, because a poor genital self-perception correlated with an impaired mental HRQoL. Since studies with non-operated men suggest that some adapt well to their penile condition, further studies should also include non-operated hypospadias.

3.2. Introduction

Hypospadias is not only operated to improve micturition, sexual function and appearance but to relieve the psychological burden of a genital malformation as well. These surgical goals should be a prerequisite for optimizing the health related quality of life (HRQoL) of affected men.

The multidimensional concept of HRQoL describes the patients' assessments of their physical, cognitive, social, and emotional functioning. A study by Schonbucher et al. (2008a) on HRQoL of children and adolescents (7-17 years old) after hypospadias repair showed that hypospadias patients at that age reported a lower HRQoL in most dimensions. However, higher age at examination was a predictor of a better HRQoL. A study by Jones et al. (2009) on adolescents with corrected hypospadias (13-15 years old) found a quality of life that was comparable to published data on "normal" children. So far, there have been no investigations that assessed the HRQoL of adults with a corrected, isolated hypospadias by means of a validated questionnaire. Nevertheless, their HRQoL may be impaired, e.g. as a consequence of a negative genital self-perception that was found particularly in adults with proximal hypospadias (Mondaini, Ponchietti, Bonafè, et al., 2002; Mureau, Slijper, van der Meulen, et al., 1995). Furthermore, other potential risk factors for HRQoL, such as erectile dysfunction, ejaculation difficulties (Rynja, de Jong, et al., 2011) and a lower orgasmic function (Rynja et al., 2009), were reported more often by patients with operated hypospadias compared to different groups of control subjects, some of them age-matched healthy controls. The only study on HRQoL of adult hypospadias patients examined severe hypospadias with genital ambiguity (Miller & Grant, 1997). In this study, the majority of men classified their general health as quite good. Nevertheless, some reported that their quality of life and their psychological well-being were impaired because of their urological condition.

The aim of the present study was to evaluate the HRQoL of men with a corrected hypospadias in comparison with non-affected, circumcised controls. Furthermore, the most relevant determinants of hypospadias patients' HRQoL were examined.

Erectile dysfunction, ejaculation difficulties and a more negative genital self-perception may be risk factors for an impaired HRQoL and were reported to be more frequent in hypospadias patients (Mondaini, Ponchietti, Bonafè, et al., 2002; Mureau, Slijper, van der Meulen, et al., 1995; Rynja, de Jong, et al., 2011). Therefore, we hypothesized that men with a corrected hypospadias have an impaired HRQoL. Furthermore, we assumed that "being in an intimate heterosexual or homosexual relationship", increasing age (older patients may have adapted to their penile conditions) and a mild type of hypospadias were predictors of a better HRQoL.

3.3. Methods

3.3.1. Subjects

Questionnaires were sent to a random sample of 218 hypospadias patients and 328 circumcised men as controls. Because an operated penis of a hypospadias patient looks similar to a circumcised one (when not operated with foreskin sparing techniques), circumcised men were chosen as controls to measure the pure effect of hypospadias.

All participants were older than 18 years and had been treated in their childhood at the University Children's Hospital, with the exception of 1 hypospadias patient and 3 circumcised men who had been operated elsewhere.

3.3.2. Main outcome measures

Health-related quality of life (HRQoL)

The Medical Outcomes Study Short Form-36 item questionnaire (SF-36) (Ware et al., 2000) assesses HRQoL in adults and contains eight subscales: physical function, role limitations due to physical problems, bodily pain, general health perception, mental health, role limitations due to emotional problems, vitality, and social function. Scores of each domain ranges from 0 to 100, with higher scores relating to better HRQoL. The physical (PCS) and the mental (MCS) component summary score are computed by summarizing relevant domains and are standardized with a mean of 50 (SD=10). In the present survey, the German version was used, and the HRQoL of the preceding 4 weeks before completing the questionnaire was investigated (Bullinger & Kirchberger, 1998). This is the common time frame used in quality of life studies among individuals with chronic conditions or malformations. Psychometric properties of the SF-36 have been reported to be excellent (Ware et al., 2000).

Genital self-perception (PPS)

Participants were asked to assess their own genitals with the Penile Perception Score (PPS), a validated instrument to assess penile self-perception after hypospadias repair and to evaluate the surgical outcome by uninvolved urologists (Weber et al., 2013). The instrument consists of the following four items to evaluate the satisfaction with various aspects of the penis on a 4-point scale ranging from very dissatisfied (0) to very satisfied (3): Position and shape of the meatus, shape of the glans, shape of the shaft skin, and general appearance of the penis. The PPS is the sum of these 4 items, ranging from 0-12. It has a good stability, a fair inter-rater

reliability and a good inter-rater reliability for ranks. In the present sample of hypospadias patients, the internal consistency of the PPS total score was good (Cronbach's Alpha: .77).

Erectile Function and Orgasmic Function

Erectile function (EF) and orgasmic function (OF) were assessed with the respective subscales of the International Index of Erectile Function (IIEF) (Rosen et al., 1997), a validated and reliable 15-item questionnaire that measures male sexual function in five separate domains.The orgasmic function subscale (OF) assesses the frequency of ejaculation and feeling of an orgasm after sexual stimulation or intercourse. The erectile function subscale (EF) measures erection frequency, erection firmness, penetration ability, maintenance frequency, maintenance ability, and erection confidence. Maximum domain score is 10 for orgasmic function (OF), and 30 for erectile function (EF), with higher scores corresponding to better orgasmic function (OF), and better erectile function (EF), respectively. In the present sample of hypospadias patients, the internal consistency of both the OF and the EF scale was good (Cronbach's Alpha OF = .90, EF = .76).

Socioeconomic status (SES)

Socioeconomic status (SES) for adults was calculated on the basis of their education and occupation and both were scored on a 6-point scale. SES scores (range: 2-12 points) were subdivided into three social classes: lower social class (2-5 points); middle social class (6-9 points) and upper social class (10-12 points). This measure proved to be a reliable and valid indicator of SES when previously used in our community (Landolt et al., 2009).

3.3.3. Procedure

After approval by the local research ethical committee, the questionnaires were sent to all participants, who were asked to return the completed questionnaires and a signed consent form.

3.3.4. Statistical analysis

Data were analyzed with the statistical program SPSS (Version 16) for Windows. T-tests were used to compare groups on continuous data. Because most variables were ordinal or categorical, nonparametric statistics were applied if available (Spearman-Brown rank

correlations, Mann-Whitney *U* tests or χ^2 -Tests). A $p < 0.05$ was considered significant. Effect sizes (Cohen's d) were calculated to show clinical relevance of group differences. Two multiple regressions were computed. Dependent variables were physical component summary score (PCS) and mental component summary score (MCS). According to their statistical importance in the bivariate analysis, and based on previous findings, the following predictors were chosen: age, living in a relationship, severity of hypospadias, genital self-perception (PPS), orgasmic function (OF), and erectile function (EF).

3.4. Results

3.4.1. Characteristics of the study sample

Of all contacted hypospadias patients and controls, 11 patients and 21 circumcised men could not be reached. Fifty-two (25.1%) hypospadias patients and 46 (15.0 %) circumcised men returned a completed SF-36 questionnaire. Seven hypospadias patients who completed the SF-36 questionnaire were excluded for one of the following exclusion criteria: A condition that affect the physical or mental health of an individual and cannot be corrected or had not been corrected until this study was performed, persistent uncorrected hypospadias or other findings for an intersex disorder besides hypospadias.

Table 8 shows the demographic and sexual characteristics of the final study sample (n=91), consisting of 45 hypospadias patients (mean-age: 26.2 years +/- 5.1) and 46 controls (mean-age: 25.5 years +/- 4.9) with an age range of 18 to 41 years. No significant differences were observed in demographic characteristics between hypospadias patients and control subjects with regard to age, nationality, SES and relationship status. In addition, hypospadias patients reported the same erectile function (EF) and orgasmic function (OF) but a significantly more negative genital self-perception compared to their controls. According to the erectile dysfunction classification (ED) of Cappelleri, Rosen, Smith, Mishra, and Osterloh (1999), no hypospadias patient had a severe or complete ED, only a few (n=11) had a mild or moderate ED, and most of them (n=26) had no such dysfunction.

All control subjects were circumcised because of phimosis at a mean-age of 6.15 years (SD=3.19; 0.42 -14.00). No complications occurred in this control group.

Twenty-two of the hypospadias patients had proximal hypospadias and 23 distal hypospadias. Hypospadias patients were operated between 1976 and 1998 at a mean age of 4.17 years for the first operation (SD=2.65; range 0.25-11.08). The last operation was performed when the patients were on average 5.23 years (SD=3.82; range 0.25-23.75). They were operated using the following techniques, of which, due to the retrospective design of the study, some are no

longer used today: MAGPI procedure (n=11), Denis Brown technique (n=5), Mathieu`s repair (n=12), island flap techniques (n=5) and various other techniques (n= 12). Most of the patients (n=31) underwent one procedure, whereas 14 were operated twice or more (mean +/- SD= 1.42 +/-0.72), mostly due to complications such as fistulas or stenosis.

Table 8: Demographic and sexual characteristics of patients and control subjects

	Hypospadias patients (n=45)		Circumcised men (n=46)		p†
Age					
Mean (SD)	26.18	(5.07)	25.52	(4.88)	0.53
Median (range)	26	(18-41)	25	(18-41)	
Nationality (n)					
Swiss	41	(91.1%)	42	(91.3%)	1.00
Foreigners	3	(6.7%)	4	(8.7%)	
Unknown	1	(2.2%)	0	(0.0%)	
Socioeconomic status (n)					
Lower	0	(0.0%)	1	(2.2%)	0.52 °
Middle	23	(51.1%)	21	(45.7%)	
Upper	16	(35.6%)	19	(41.3%)	
Unknown	6	(13.3%)	5	(10.8%)	
Relationship status (n)					
Living in a relationship	22	(48.9%)	31	(67.4%)	0.09
Single	22	(48.9%)	14	(30.4%)	
Unknown	1	(2.2%)	1	(2.2%)	
PPS					
Mean (SD)	8.23	(2.06)	9.73	(1.80)	**0.002**
Median (range)	8	(3-12)	10	(6-12)	
OF					
Mean (SD)	8.95	(2.43)	8.73	(2.79)	0.71
Median (range)	10	(0-10)	10	(0-10)	
EF*					
Mean (SD)	26.03	(6.00)	24.44	(7.26)	0.26
Median (range)	29	(13-30)	28	(2-30)	
No dysfunction (EF score 26 to 30)	26	(57.8%)	27	(58.7%)	
Mild dysfunction (EF score 22 to 25)	4	(8.9%)	1	(2.2%)	
Mild to moderate dysfunction (EF score 17 to 21)	0	(0.0%)	2	(4.3%)	
Moderate dysfunction (EF score 11 to 16)	7	(15.6%)	7	(15.2%)	
Severe dysfunction (EF score 6 to 10)	0	(0.0%)	1	(2.2%)	
Unknown	8	(17.7%)	8	(17.4%)	

† T-tests for continuous data; U test according to Mann-Whitney for ordinal data; χ^2-tests for categorical variables.
° U test according to Mann-Whitney on the basis of SES scores.
* Erectile dysfunction classification according to Cappelleri et al. (1999).

3.4.2. Health-related quality of life (HRQoL)

As displayed in Table 9, no significant differences between hypospadias patients and controls were found in any of the SF-36 subscales, the physical component summary score (PCS) or the mental component summary score (MCS). Effect sizes were small.

Furthermore, U tests according to Mann-Whitney showed no significant differences in the physical and the mental dimensions of HRQoL between hypospadias patients and controls (p=.69 for PCS and p=.25 for MCS) when comparing patients with proximal hypospadias (mean= 55.84, SD=6.04 for PCS and mean= 52.31, SD=9.65 for MCS) separately with circumcised men (mean=56.60, SD=2.83 for PCS and mean=50.38, SD=7.83 for MCS). Likewise, there were no significant differences in the physical and the mental dimensions of HRQoL (p=.67 for PCS and p=.51 for MCS) when comparing patients with distal hypospadias (mean=55.61, SD=4.77 for PCS and mean=52.59, SD=7.01 for MCS) separately with circumcised men (mean=56.60, SD=2.83 for PCS and mean=50.38, SD=7.83 for MCS). Also, the other subscales of the SF-36 did not significantly differ between patients with distal hypospadias and controls respectively between patients with proximal hypospadias and controls (data not shown). Effect sizes for all subscales and component summary scores were small (Cohen's d <.50).

In addition, results showed that there were no significant differences in HRQoL between hypospadias patients and controls when comparing patients with "one" or "more than one operation" separately with circumcised men (Table 10 & Table 11).

Table 9: Hypospadias patients' health-related quality of life (HRQoL): Comparison with control subjects

	Hypospadias Patients (n=45)		**Circumcised men (n=46)**			
SF-36 scales †	**Mean**	**SD**	**Mean**	**SD**	**Effect size d**	**p***
Physical function	96.33	15.17	97.50	8.48	-0.10	0.90
Role limitations due to physical problems	95.56	14.39	97.28	9.47	-0.14	0.68
Bodily pain	91.86	18.80	94.35	11.07	-0.16	0.92
General health perception	81.77	14.38	78.59	14.65	0.22	0.27
Mental health	80.36	13.50	76.96	15.04	0.24	0.30
Role limitations due to emotional problems	91.85	21.50	91.30	20.41	0.03	0.82
Vitality	66.70	15.81	65.33	15.65	0.09	0.62
Social function	93.75	15.60	89.67	18.12	0.24	0.17
Physical component summary score (PCS)	55.72	5.35	56.60	2.83	-0.21	0.97
Mental component summary score (MCS)	52.46	8.28	50.38	7.83	0.26	0.27

Effect sizes according to Cohen: 0.20 small effect size, 0.50 medium effect size, >0.80 large effect size.

***** U test according to Mann-Whitney.

† Higher scores indicate better HRQoL.

Table 10: Health-related quality of life among hypospadias patients with one operation compared to circumcised men

	Hypospadias patients with one operation (n=31)		Circumcised men (n=46)			
SF-36 scales †	**Mean**	**SD**	**Mean**	**SD**	**Effect size d**	**p***
Physical function	95.48	18.18	97.50	8.48	-0.14	0.64
Role limitations due to physical problems	96.77	10.69	97.28	9.47	-0.05	0.88
Bodily pain	93.48	14.54	94.35	11.07	-0.07	0.80
General health perception	81.45	15.02	78.59	14.65	0.19	0.40
Mental health	81.42	10.66	76.96	15.04	0.34	0.29
Role limitations due to emotional problems	93.55	15.91	91.30	20.41	0.12	0.81
Vitality	66.94	14.36	65.33	15.65	0.11	0.76
Social function	93.95	13.26	89.67	18.12	0.27	0.31
Physical component summary score (PCS)	55.74	4.85	56.60	2.83	-0.22	0.90
Mental component summary score (MCS)	52.81	6.44	50.38	7.83	0.34	0.36

Effect sizes according to Cohen: 0.20 small effect size, 0.50 medium effect size, >0.80 large effect size.

* U test according to Mann-Whitney.

† Higher scores indicate better HRQoL.

Table 11: Health-related quality of life among hypospadias patients with more than one operation compared to circumcised men

	Hypospadias patients with more than one operation (n=14)		Circumcised men (n=46)			
SF-36 scales †	**Mean**	**SD**	**Mean**	**SD**	**Effect size d**	**p***
Physical function	98.21	3.17	97.50	8.48	0.11	0.64
Role limitations due to physical problems	92.86	20.64	97.28	9.47	-0.28	0.51
Bodily pain	88.00	26.73	94.35	11.07	-0.31	0.84
General health perception	82.54	13.28	78.59	14.65	0.28	0.32
Mental health	77.85	18.95	76.96	15.04	0.05	0.63
Role limitations due to emotional problems	88.10	30.96	91.30	20.41	-0.12	0.91
Vitality	66.15	19.49	65.33	15.65	0.05	0.55
Social function	93.27	20.80	89.67	18.12	0.18	0.20
Physical component summary score (PCS)	55.66	6.63	56.60	2.83	-0.18	0.88
Mental component summary score (MCS)	51.63	11.87	50.38	7.83	0.12	0.36

Effect sizes according to Cohen: 0.20 small effect size, 0.50 medium effect size, >0.80 large effect size.

* U test according to Mann-Whitney.

† Higher scores indicate better HRQoL.

3.4.3. Determinants of health-related quality of life (HRQoL) of hypospadias patients

Results of the multiple regression analyses are shown in Table 12. While no variable was significantly associated with better physical HRQoL (PCS), a more positive genital self-perception (PPS) and a better orgasmic function (OF) were significantly associated with a better mental HRQoL. The independent variables accounted for -2% (PCS) and 51% (MCS) of the variances of the dependent variables; the regression model for predicting mental HRQoL was significant, whereas the regression model for predicting physical HRQoL was not significant.

A further calculation showed that no medical variable correlated with the physical or mental HRQoL: Correlations with the physical HRQoL (PCS) were (r= -.01; p= .97) for age at first operation, (r= .05; p= .74) for age at last operation and (r= -.02; p= .90) for number of operations. Correlations with the mental HRQoL (MCS) were (r= .05; p= .73) for age at first operation, (r= .04; p= .80) for age at last operation and (r= .03; p= .83) for number of operations.

Table 12: Multiple regression analyses for predictors of physical health-related quality of life (PCS) and mental health-related quality of life (MCS) of hypospadias patients

Equation 1: ***physical health-related quality of life (PCS)***					**Equation 2:** ***mental health-related quality of life (MCS)***				
Variable	**B**	**SEB**	**Beta**	**p**	**Variable**	**B**	**SEB**	**Beta**	**p**
Age	0.23	0.18	0.25	0.20	**Age**	0.17	0.24	0.09	0.48
Living in a relationship	-0.98	1.98	-0.11	0.63	**Living in a relationship**	-0.33	2.67	-0.02	0.90
Severity of hypospadias	1.25	1.72	0.14	0.47	**Severity of hypospadias**	0.38	2.32	0.02	0.87
PPS	-0.18	0.40	-0.08	0.66	**PPS**	1.34	0.54	0.32	**0.020***
OF	0.05	0.39	0.02	0.91	**OF**	2.13	0.52	0.58	**0.000****
EF	-0.11	0.17	-0.15	0.51	**EF**	0.16	0.23	0.11	0.50

Equation 1: $R = 0.40$ (n =35, $F = 0.91$, $p = 0.50$), $R^2 = 0.16$, R^2 adj = -0.02 **< 0.05, **< 0.01*

Equation 2: $R = 0.77$ (n =35, $F = 6.80$, $\mathbf{p = 0.000}$), $R^2 = 0.59$, R^2 adj = 0.51 **< 0.05, **< 0.01*

3.5. Discussion

This cross-sectional study is the first to have investigated the HRQoL of adult hypospadias patients. In contrast to our hypothesis, adults with corrected hypospadias reported a similar HRQoL to that of a control group of circumcised men.

These results are in line with those of Jones et al. (2009), who investigated a sample of adolescents. Schonbucher et al. (2008a) showed a poorer HRQoL of children and adolescents with corrected hypospadias when compared to controls. However, their results showed that HRQoL improved with increasing age. Even in the study by Miller and Grant (1997), the majority of men with severe hypospadias classified their general health as quite good, although some reported that both quality of life and psychological well-being were impaired. However, in contrast to our study, all participants of that study had a severe hypospadias with genital ambiguity. It seems that the HRQoL changes with increasing age: While children with corrected hypospadias have a poorer HRQoL, adolescents already have a better HRQoL, and finally, as shown in our study, it seems that adults have the same HRQoL as the control group.

Multiple factors may explain why, in our study, adults with corrected hypospadias have a normal HRQoL. Their learning to deal with their corrected hypospadias and some appropriate coping styles may have improved their HRQoL over time. Furthermore, possible traumatic memories of the treatment or complications in childhood date back many years and may have been mastered.

The present study also investigated predictors of physical and mental dimensions of HRQoL. In contrast to our hypothesis, none of the variables examined turned out to be a predictor of *physical* dimensions of HRQoL. However, the *mental* dimensions of HRQoL were impaired by a worse genital self-perception and a lower orgasmic function. Results concerning genital self-perception are in line with the study by Schonbucher et al. (2008a), who found a significant positive correlation between mother-reported child HRQoL and penile self-perception in older children. Furthermore, our results are congruent with those from studies on psychosocial adjustment, a topic that is related to mental dimensions of HRQoL. Those studies (Mureau, Slijper, Slob, & Verhulst, 1997) indicated that a better genital self-perception was associated with a better psychosocial functioning. However, correlations were small, and the significant ones were found for younger hypospadias patients (children and adolescents).

An unexpected finding of our study was that age, relationship status, and severity of hypospadias did not emerge as predictors for both physical and mental dimensions of

HRQoL. Astonishingly, not even erectile function, when measured with the International Index of Erectile Function (IIEF) (Rosen et al., 1997), was a predictor of any dimension of HRQoL, although recent studies (Rynja, de Jong, et al., 2011) have shown that erectile dysfunction is more prevalent in hypospadias patients compared to controls. Since our sample of hypospadias patients had a good erectile function, comparable to that of circumcised men, and no one reported a severe or complete erectile dysfunction, it is possible that this variable would be a predictor of HRQoL in a more representative sample of individuals with hypospadias. Furthermore, neither age at first operation, age at last operation, nor number of operations were associated with the physical or mental dimensions of HRQoL. This again is perfectly in line with the findings of Schonbucher et al. (2008a), who also showed that severity of hypospadias was not a risk factor for HRQoL in children and adolescents and that neither the age at first operation, the number of surgeries nor any other medical variable correlated with HRQoL. Furthermore, our results correspond with those from studies on psychological adjustment showing that medical characteristics were not related to psychological adjustment of men with corrected hypospadias (Berg, Berg, & Svensson, 1982; Mureau et al., 1997). Theories on psychological development (Schultz, Klykylo, & Wacksman, 1983), the personal experience of many surgeons (Manzoni et al., 2004) as well as the American Academy of Pediatrics (American Academy of Pediatrics, 1996) suggest that operations around the first birthday should have the smallest negative influence on HRQoL, although these notions lack evidence. In accordance with the pilot study by Weber et al. (2009)**,** we could not find any significant correlation between age at operation and HRQoL.

Limitations

Although this is the first study to assess the HRQoL of adult patients with isolated hypospadias by means of a standardized and validated questionnaire, some limitations need to be addressed. First, because the participation rate was rather low, it is possible that most of the study participants were individuals who had a positive approach to their corrected hypospadias and for whom the issue was not a taboo. This could have biased our results towards a too positive HRQoL.

Second, a post-hoc power analysis ($\alpha = .05$; two-tailed) using the G*power software by Faul, Erdfelder, Lang, and Buchner (2007), indicated that the study is of limited statistical power. For each U-Test according to Mann-Whitney, the power to detect a large effect size (d= .80) was adequate (.97) but the power to detect a medium (d= .50) or small effect size (d= .20) was below the recommended .80 level (Cohen, 1988)**.** For the two regression analyses, the power to reveal a large ($f^2 = .35$), medium ($f^2 = .15$) and small effect size ($f^2 = .02$) was less than

adequate. Therefore, it is not excluded that with a larger sample size results would be different.

Third, it is possible that by using a standardized and validated generic HRQoL-questionnaire some problems of men with corrected hypospadias were not appropriately addressed (e.g., sexual activity). Therefore, the development of a disease-specific HRQoL measure for hypospadias patients should be taken into consideration (Schonbucher et al., 2008a).

Fourth, we should consider that circumcision is not very common in Switzerland. As the HRQoL of our study sample was compared with that of circumcised men, the current results do not allow generalization to non-circumcised men. In a future study, it is therefore important to compare the HRQoL of men with corrected hypospadias with that of non-affected, non-circumcised men. In addition, because in the present sample of hypospadias patients only 3 men were non-Swiss, and only 2 men had a different religious background (Muslim), our findings can not be easily generalized to other cultural groups.

Fifth, the hypospadias patients in this cohort were, according to today's standards, operated at a rather late mean age of 4.17 years, and only 6 patients were operated before 18 months. This is due to the fact, that the study was performed on adults who were operated between 1976 and 1998. Although age at the first operation did not correlate with the HRQoL, no conclusions should be drawn from these limited numbers and future studies with a cohort of patients operated at a younger age are needed to clarify this aspect.

Since a large proportion of the variance (especially that concerning the physical dimensions of HRQoL) could not be explained by the variables examined, there must be other predictors.

Referring to studies on long-term outcome of hypospadias repair (Rynja, de Jong, et al., 2011), albeit with methodological problems, the following outcome-variables may be of particular importance for HRQoL: Micturition (e.g. satisfaction with urinary stream), further aspects of sexuality (e.g. sexual satisfaction) or further aspects of genital perception (e.g. satisfaction with penile size). Moreover, based on results in children and adolescents (Mureau, Slijper, Nijman, et al., 1995; Schonbucher et al., 2008a) it is possible that being ashamed of the penile appearance or being teased and receiving adverse comments concerning genital appearance are also risk factors of an impaired HRQoL of adult hypospadias patients. Finally, the parents' attitude towards hypospadias may have a profound influence on the child's psychological adjustment as suggested by Easson (1966) because the emotional reaction of them might be transmitted to their children and may therefore affect their children's feelings.

Clinical Implications

This study showed genital self-perception and orgasmic function to be of particular importance for mental dimensions of HRQoL. Therefore, to prevent impaired mental HRQoL, patients with hypospadias should be supported in developing a positive genital self-perception and a satisfactory orgasmic function. With respect to the latter, we have to consider that more recent surgical techniques than those applied to this cohort of patients, who were operated up to 36 years ago, may lead to better surgical results and may in consequence have less negative impact on HRQoL. However, even an objectively good cosmetic result must not be associated with a favourable genital self-perception. It is quite possible that even nowadays, patients with a good surgical result are still afraid of adverse comments due to their penile appearance. A similar phenomenon is the frequently expressed concern of healthy men regarding their penile length, although their penis is normally sized (Mondaini, Ponchietti, Gontero, et al., 2002). The development of a positive genital self-perception can possibly be supported by informing patients and parents comprehensively about the hypospadias and by giving them longer follow-up care. We support a late follow-up in adolescence, as suggested by Bracka (1989) and we believe that informative discussions between surgeons, parents and patients are as crucial as taking an informed consent. In an initial period, families must become aware that their own attitude towards the malformation may influence their child's psychological development profoundly (Easson, 1966). Therefore, preoperative counselling must not only include information on etiology, genetics, fertility issues and surgical techniques but also provide new insights into psychological aspects. These aspects must be rediscussed during follow-up visits and must also be reviewed with the patient during a follow-up visit in adolescence. If urologists are in doubt regarding a patient's well-being, they may recommend psychological assessment and treatment, as suggested by Schonbucher et al. (2008a).

There is some evidence that longer follow-up care and a comprehensive briefing are important. Specifically, in the study by Bracka (1989), most patients reported that they were not guided adequately and had never heard about hypospadias. This contributed to avoiding the use of public showers for fear of being ridiculed or rejected by others because of the corrected hypospadias. Furthermore, in the study by Mondaini, Ponchietti, Bonafè, et al. (2002), most hypospadias patients reported that their embarrassment was relieved when they were informed about several aspects of hypospadias (e.g. incidence, future sexuality etc.). Moreover, the study by Aho et al. (1997) showed that patients who would have preferred a longer follow-up were more unsatisfied with the surgical result or with the penile appearance.

As Schonbucher et al. (2008a) have mentioned, it is important not only to assess the surgical outcome after a correction, but also to measure the HRQoL of the patients. In doing so, it is important to interview both patients and parents, because their valuations can differ

considerably. In addition, a psychological treatment for a patient (at least for severe types of hypospadias) may help.

Because our study showed that hypospadias patients have an inferior genital self-perception compared to controls, and since a negative genital self-perception is a risk factor for impaired mental dimensions of HRQoL, an important aim of such a psychological treatment might be to establish a good acceptance of the penile appearance and to develop positive feelings toward the genitalia. However, it is not clear whether the corrected hypospadias, or perhaps even the surgery itself, is the cause of the more negative genital self-perception. It is possible that a surgical correction leads to closer attention of the patients and their parents to the hypospadias, which may support the development of a negative genital perception. Since studies (Dodds et al., 2008; Fichtner et al., 1995) on men with non-operated hypospadias have shown that many of the interviewed men and even their sexual partners (Fichtner et al., 1995) were not aware of the hypospadias, it would be important not only to assess the HRQoL of adults with corrected hypospadias in comparison with healthy, non-affected men (in order to replicate the present results with a more representative sample), but also to compare them with non-operated hypospadias patients. Perhaps such studies would demonstrate that, at least for mild types of hypospadias without functional impairment, the potential role of surgery, if any, should be discussed.

In conclusion, our data suggest that the HRQoL of adult hypospadias patients is comparable to that of a control group of circumcised men. Considering the importance of a positive genital self-perception on HRQoL, patients should be supported in with regard to this issue. Neither socio-demographic and medical variables nor erectile function turned out to be predictors of the physical or mental HRQoL. Since studies on men with non-operated hypospadias suggest that they adapt well to their penile condition, it is important for further studies not only to assess the HRQoL of adults with corrected hypospadias but to include men with non-operated hypospadias to determine which types of hypospadias actually benefit from a surgical correction.

Part C General Discussion

The main objective of our research project was to learn more about the long-term psychological outcome of hypospadias repair. These results should serve to generate conclusions for improved therapy of children with hypospadias.

This issue was examined in two steps. In the first step, we wanted to know how laypersons unacquainted with hypospadias evaluate the cosmetic result of HASRGs of adults. Thus, we wanted to know whether they notice a difference between HASRGs and non-affected, circumcised genitals and whether they perceive HASRGs as normal in appearance. In this study, we also sought to identify the characteristics of perceivers that predict a more positive perception of HASRGs. To learn more about genital perception by others, we also investigated how relevant women considered single aspects of a penis in general. The study design and the results were presented in section B1 and B2. In a second step, we wanted to know whether men with corrected hypospadias adapt to their penile condition, that is, if they have normal HRQoL compared to non-affected, circumcised men. In addition, we wanted to know which variables are determinants of HRQoL in men with corrected hypospadias. The design and the results of this study were presented in section B3.

This chapter summarizes and discusses the main findings of this research project (section C1). In addition, the strengths and limitations of the present research project and conclusions for future research activities are considered (section C2). Finally, some suggestions for clinical practice (section C3) and a general conclusion of study A and B (section C4) are outlined.

1. Summary and general reflections on findings

1.1. Summary and general reflections on the findings of study A:

1.1.1. Genital perception of hypospadias-affected surgically-repaired genitals by laypersons

Our research project A was the first to quantitatively assess the following research questions:

Do laypersons unacquainted with hypospadias notice a difference between HASRGs and non-affected genitals? Do female laypersons perceive HASRGs as normal-looking? Which perceiver-related characteristics are predictors of a more favorable laypersons' genital perception of HASRGs? And, finally, which aspects of a penis are considered relevant for a satisfactory penile appearance?

It was hypothesized that laypersons unacquainted with hypospadias do not notice a difference between HASRGs and circumcised genitals. In line with this hypothesis, we further expected female laypersons to perceive HASRGs as being as normal-looking as the genitals of non-affected, circumcised genitals.

At first sight, the results of the present research project seemed to be inconsistent with our hypotheses: HASRGs were perceived as slightly, but significantly less positive and as less normal-looking than circumcised genitals. However, when the genitals of distal and more proximal types of hypospadias were regarded separately, only genitals with more proximal types of hypospadias were rated as inferior and as less normal-looking than circumcised genitals. In line with our hypotheses, genitals with distal hypospadias were perceived as normal-looking and as positively as circumcised genitals. Fortunately, the majority of hypospadias patients have a mild type of hypospadias. And that means that, according to our results, the majority of patients do not need to worry about negative comments from peers or potential sexual partners. Interestingly, our results also indicated that even patients with very uncommon, more proximal types of hypospadias probably do not need to worry about the negative reactions of the social environment. According to our calculations of effect sizes, the perceived difference between the genitals of patients with more proximal type of hypospadias and of circumcised men cannot be considered clinically relevant, as the difference and therefore the effect size were small. In absolute numbers, the mean difference between more proximal forms of HASRGs and circumcised genitals was only 0.13 on a 4 point-Likert scale regarding the mean score of laypersons' genital perception and 0.24 on a 4-point Likert scale regarding the question whether an illustrated HASRG penis appeared normal.

Interestingly, the average rating of both the genitals of men with corrected hypospadias and those of circumcised men was something between "unsatisfactory" and "satisfactory" or something between "rather abnormal" and "rather normal". Hence, there seem to be other factors which are not hypospadias-related (such as whether the pubic hair is shaved or not) that possibly play an equally important role for the evaluation of the penile appearance of HASRGs as hypospadias-related penile aspects such as the shape and position of the neo-meatus. However, as circumcision is not very common in Switzerland, the absence of the foreskin may have caused the generally more negative evaluation of patients' and controls' genitals.

The rather negative evaluation of both HASRGs and circumcised genitals is in line with several studies on genital self-perception of hypospadias patients and non-affected men (Rynja, de Jong, et al., 2011). They showed that not only men with corrected hypospadias but also non-affected controls suppose that their penile appearance differs from normal. There seems to be general uncertainty about what a so-called "normal" penis looks like.

Our result that severe, penile and proximal forms of HASRGs were rated less positively than milder, distal HASRGs seems to meet patients' expectations. Previous studies have shown that a lower genital self-perception was associated with more proximal hypospadias (Chertin et al., 2013; Liu et al., 2006; Mureau, Slijper, van der Meulen, et al., 1995; Mureau, Slijper, Slob, et al., 1995; Rynja, de Jong, et al., 2011; Rynja et al., 2012). Furthermore, men with proximal hypospadias reported hearing silly remarks or being ridiculed because of their penile appearance more often than those with distal forms (Liu et al., 2006). In contrast, a review of existing studies (Rynja, de Jong, et al., 2011) showed that concern about an abnormal penile appearance is less often mentioned by men with a severe type of hypospadias than controls. However, the reviewing authors observed that this remarkable difference could be caused by the age difference between the patients and controls. They conclude that the expectation of what a penis "normally" looks like is gained with increasing age and is influenced by cultural concepts and by sexual experience.

Various considerations suggest that in reality laypersons' evaluations of HASRGs would be even more favorable than in the experimental setting of the study. First, we have to consider that laypersons' rating was only based on photographed genitals without knowing anything about the individuals photographed. It is quite obvious that in actual situations, such as when a couple interacts sexually, various factors such as a patient's general physical attractiveness and how well the individuals involved know each other may influence the perception of a

patient's penis. Second, people do not tend to observe their partners' penis closely and they do not compare it with other genitals at the same time, as in the experimental-setting.

It should be also borne in mind that patients with proximal hypospadias who are treated with the most recent techniques may have a more favorable outcome than those examined in our study who have been treated a rather long time ago, so their perception by others may well be similar to that of distal hypospadias. And, although we found that laypersons perceive a difference between HASRGs and circumcised genitals, it does not automatically mean that the perceived difference matters to them. Specifically, some research suggests that the cosmetic penile appearance of men with corrected hypospadias probably does not bother their intimate partners (Maier & Tewes, 1984).

Thus, overall our study A indicates that most men with corrected hypospadias (those with mild types) do not have to be afraid of the negative reactions of others. However, as mentioned in the introduction (chapter A 2.4.1.) two older studies (Mureau, Slijper, Nijman, et al., 1995; Svensson et al., 1981) indicated that more hypospadias patients than controls had received negative reactions due to their penile appearance. However, these inconsistent results only reflect the patients' views. Analysis of the situation of men with corrected hypospadias based on Cash's CB model (Cash, 2002, 2011) may lead to the prediction that those with a negative genital self-perception tend to interpret any comment in relation to their penis in a negative way, regardless of whether the comment was formulated neutrally or was actually negative. In line with this, some scientific evidence (Rosser, Moss, & Rumsey, 2010) indicates that individuals with a high level of appearance concern interpret ambiguous stimuli as both negative and appearance-related. Moreover, a recently published study (Vandendriessche et al., 2010) found that adolescents with operated hypospadias were not ridiculed significantly more often than non-affected controls. And finally, studies on sexuality (Rynja, de Jong, et al., 2011) showed that men with corrected hypospadias did not report being sexually less active than controls. This may indicate that sexual partners do not reject men with corrected hypospadias because of their penile appearance.

Unfortunately, a tendency to interpret comments or attitudes regarding penile appearance as negative actually provokes negative reactions by others. According to the theory of self-fulfilling prophecy by Merton (1948), an expected behavior of others will happen because it is forced by one's own behavior. That means, for example, that any inhibited behavior that hypospadias patients show, for instance in public toilets, because of their negative genital self-perception may itself provoke such negative comments by peers.

However, it is not only the characteristics of the hypospadias patients, such as shyness or the type of hypospadias, but also the characteristics of the perceivers, such as sexual experience, which may influence how HASRGs may be assessed by others. In the following section, some perceiver-related characteristics are discussed which in the current study were found to be predictors of a more favorable genital perception by laypersons.

1.1.2. Determinants of laypersons' genital perception of HASRGs

Research project A also investigated which characteristics of the observing laypersons influenced their evaluation of whether penile appearance of men with corrected hypospadias is considered as satisfactory (study 1) and as normal-looking (study 2).

In study 1, we examined whether the following variables were determinants of laypersons' genital perception of HASRGs: gender of the rater, being in an intimate relationship or not, socio-economic status, age, degree of sexual interest and sexual experience, and genital self-perception of the rater. In line with our expectations, the multiple regression analysis revealed four significant predictors of a better laypersons' perception of HASRGs: higher age, being in an intimate relationship, higher socio-economic status (SES) and a higher sexual interest.

In study 2, only answers of female laypersons were taken into consideration. We examined which variables influence women's evaluation of whether a penis is considered normal. Variables examined were: age, degree of sexual interest, number of sexual partners, and whether a woman perceives the meatus, the penile size or the general penile appearance as important. As hypothesized, there were four significant predictors of perceiving a hypospadias-affected penis as more normal: Higher age, higher sexual interest, and perceiving the "general cosmetic appearance of a penis" as more important and "penile length" as less important.

Thus, in both studies, two identical variables turned out to be a predictor of considering HASRGs as more satisfactory or as more normal-looking, a higher age and a higher degree of sexual interest in the laypersons. Interestingly, they also predicted a more favorable evaluation of circumcised genitals. Thus, higher age and a higher sexual interest seem not only to be determinants of perception of malformed genitals but also of non-affected, so-called "normal" genitals.

In line with our results, some authors (Moss & Rosser, 2008) suggest that individuals may find it easier with increasing age to accept their own physical appearance. This tolerant perception of one's own body may give rise to a more open-minded evaluation of the body of

others and thus possibly also of the genital appearance of others. In addition, with increasing age and with a higher degree of sexual interest, people become more sexually experienced and know a broader range of different genital appearances. Thus, their opinion of a perfect penile appearance becomes more realistic and therefore they may be less prone to reject partners due to genital imperfections. In line with our results, a positive association between better genital perception and increasing age (Bramwell & Morland, 2009) as well as a correlation between a positive genital perception and higher sexual interest (respectively sexual desire) was also found in studies on the genital self-perception of women (Ålgars et al., 2011; Berman et al., 2003).

In our study, a better laypersons' genital perception of HASRGs was also found to be associated with being in an intimate relationship. This may be because the laypersons who have been in an intimate relationship may have developed a more realistic image of genital appearance due to their familiarity with their partners' genitals. However, we have to keep in mind that the majority of laypersons questioned in our study were in an intimate relationship, which may have biased the results.

In addition, it is conceivable that educational background, by providing laypersons with more information about malformations and variability of penile appearance, influences their perception of HASRGs in a positive way. Notably, there are no previous findings on this issue. Also, middle and upper socio-economic status were overrepresented, and thus any conclusion must be made carefully.

In contrast to our hypotheses and to studies regarding the genital self-perception of individuals without genital malformations (Morrison, Bearden, Ellis, & Harriman, 2005), sexual experience (the number of sexual partners or the fact whether laypersons have made any sexual experiences or not) did not emerge as a predictor of considering HASRGs to be either more satisfactory or more normal-looking. At first sight, these results seem to contradict our discussion above regarding the sexual experience someone gains with increasing age. However, it is likely that it depends on how long someone has dealt with the topics of sexuality and body image and not on how many sexual partners someone has had. For example, a young woman may have sexual contacts with a high number of sexual partners but only in short and casual sexual relationships, whereas an older woman may have fewer sexual partners but longer lasting intimate relationships and thus a deeper familiarity with the topics of sexuality and body image.

Studies on healthy women and men have shown that men generally have a significantly more positive genital self-perception (Morrison et al., 2004; Reinholtz & Muehlenhard, 1995) and a more positive perception of their partner's genitals (Reinholtz & Muehlenhard, 1995) than

women. Therefore, we were surprised to find that laypersons' gender, their genital self-perception and their degree of sexual experience were not associated with the outcome of their genital perception of HASRGs. However, this may be due to the relatively high sexual experience and high satisfaction with their own genitals of the participating laypersons.

In our study, we also investigated whether the rating of HASRGs is influenced by whether a woman perceives several penile aspects (such as penile size) or the general penile appearance to be important. As expected, our findings show that the less important a woman considers penile length and the more important she considers general cosmetic appearance, the more normal she evaluates photosets of HASRGs. These results are in line with Cash's CB model (Cash, 2002, 2011). The model states that cultural norms influence our opinion of what is a good-looking or normal-looking penis. Nowadays, access to pornography is very easy due to technological advances (Schick, Calabrese, Rima, & Zucker, 2010). Therefore, cultural norms concerning penile appearance may be transmitted through pornographic material. Because pornography mainly reflects an unrealistic range of penile appearance (Schick et al., 2010) such as penises of above-average sizes, people may internalize such unrealistic norms. For example, they may regard a very large penis to be average in size (Mondaini, Ponchietti, Gontero, et al., 2002). In line with this, there is scientific evidence (Morrison, Bearden, Ellis, Harriman, & Morrison, 2006) that the consumption of pornographic material correlates with the quality of genital self-perception. It can be assumed that an internalized norm concerning penile appearance not only influences the genital self-perception of individuals but also their genital perception of others. Therefore, our finding that a penis is regarded as less normal the more important a person regards penile size and as more normal the more important a person regards the whole general cosmetic appearance (instead of single penile aspects which are based on unrealistic expectations) perfectly matches the theoretical ideas discussed above.

Because the shape of the meatus is a penile aspect in which someone might recognize the existence of a corrected hypospadias, it would have been possible that HASRGs are perceived as more normal the less important a woman considers the meatus. Contrary to these expectations, the rating of the importance of the position and shape of a meatus did not correlate with the evaluation of the normality of the penile appearance. Thus, people seem to have no specific norm regarding the position and the shape of the meatus.

1.1.3. Importance of penile aspects in general

Since mild types of hypospadias (which represents the majority of hypospadias) are usually operated for cosmetic reasons, we were interested in which aspects of penile appearance are

actually evaluated as important by women. We were particularly interested in how relevant they consider the position and the shape of the meatus because these penile aspects are crucial when operating hypospadias patients. In addition, we wanted to know whether penile size is regarded as important, because more hypospadias patients (especially with severe types of hypospadias) are dissatisfied with their penile size than men without hypospadias (Mureau, Slijper, van der Meulen, et al., 1995; Rynja, de Jong, et al., 2011).

The present research showed that there is no single penile aspect which is essential for the evaluation of penile appearance. Women considered the general cosmetic appearance as most important, while all other penile aspects were regarded as clearly less relevant.

Although mild types of hypospadias are usually operated to achieve a slit-like meatus at the tip of the glans, participating women rated the position and shape of the meatus as an "unimportant" aspect of penile appearance, which was in line with our hypothesis. Compared to the other penile aspects, they also considered the meatus to be the least important aspect of penile appearance. It would have been conceivable that the position and shape of the meatus was seen as more important in comparison to the other aspects after women had seen photos of HASRGs. But even after exposure to photos of HASRGs, and thus to a non-perfect meatus, the position and shape of the meatus was still rated as "unimportant" and as the least important item of all the penile aspects. However, the women participating in our study were not exposed to photos of non-corrected hypospadias. It has also been shown indirectly in other studies that the position and shape of the meatus is perceived as less important than other penile aspects and that women rather look at the overall impression of penile appearance. For example, studies among men with non-operated hypospadias showed that many of the men interviewed (Dodds et al., 2008; Fichtner et al., 1995) and even their sexual partners (Fichtner et al., 1995) were not aware of the hypospadias even though the meatus was not at the tip of the glans. The position and shape of the meatus seems to be rather unimportant even for some patients with corrected hypospadias (Glassman et al., 1980; Kiss et al., 2011; Sommerlad, 1975). For example, one study involving men with corrected hypospadias (Kiss et al., 2011) found that satisfaction with the surgical result did not correlate with the meatus distance from penile tip but with the overall genital self-perception. Another study (Glassman et al., 1980) found no reaction or dissatisfaction of men with corrected hypospadias in relation to the positioning of their meatus, even though the meatus was not at the tip of the glans. In line with Glassman et al. (1980) we therefore suggest that not only people without any genital malformation but also hypospadias patients may regard a different positioning of the meatus to be a variation of normal.

Regarding penile size, we found that, in line with our hypothesis, penile length was not considered as very relevant by women. This is interesting, as men in general (Mondaini, Ponchietti, Gontero, et al., 2002) and especially men with a corrected hypospadias (Mureau, Slijper, van der Meulen, et al., 1995; Rynja, de Jong, et al., 2011) are often concerned about their penile size. These results are in line with Cash's CB model (Cash, 2002, 2011). As mentioned above, there seems to exist a cultural norm (transmitted partly through pornographic material) that a good-looking penis is of long size. Because more men than women were found to view pornography regularly (Zillich, 2011), mainly men seem to have internalized this cultural norm. Therefore it makes sense that men often tend to underestimate their penis size or want to have a larger penis, whereas most women find penile size "unimportant" or even "totally unimportant" (Francken et al., 2002) and reported being satisfied with their partners' penile size (Lever et al., 2006).

Overall, our findings indicated that women seem to have no particular concept of what a penis should look like. Instead of single penile aspects (like the meatus or penile size) the overall penile impression seems to influence their rating of penile appearance.

1.2. Summary and general reflections on the findings of study B:

1.2.1. Health-related quality of life in men living with corrected hypospadias

Study B is the first study to investigate the HRQoL of men with an isolated hypospadias in comparison to a non-affected control group. In contrast to our hypothesis, adults with corrected hypospadias reported similar HRQoL to that of a control group of circumcised men.

These results are in line with the few existing studies on HRQoL of men with corrected hypospadias. Namely, Chertin et al. (2013) found that men with corrected hypospadias reported physical and mental HRQoL that was within the normal range. Even in the study by Miller and Grant (1997), the majority of men with severe hypospadias classified their general health as quite good, although some reported that both quality of life and psychological well-being were impaired. However, in contrast to our study, all participants of that study had severe hypospadias with genital ambiguity. In line with our results, a study with younger adolescent hypospadias patients (Jones et al., 2009) also found a quality of life that was comparable to published data on "normal" children. In contrast, Schonbucher et al. (2008a) found impaired HRQoL of children and adolescents with corrected hypospadias when compared to controls. However, their results showed that HRQoL improved with increasing age. Therefore it seems that the HRQoL of individuals with corrected hypospadias normalizes

over time; while children have the most impaired HRQoL, adolescents already have a better HRQoL, and adults finally seem to have a HRQoL that is comparable to men without a genital malformation.

There are several possible explanations why adults with corrected hypospadias report normal HRQoL in our and in the study by Chertin et al. (2013), whereas younger individuals seem to have impaired HRQoL. First, in line with some authors (Thompson & Kent, 2001), we suggest that the simple passage of time helps adaptation to the penile condition. Second, possible traumatic memories of the treatment or complications in childhood recede over time and may be mastered. Third, higher HRQoL in adults than in children and adolescents with corrected hypospadias may be explained by the theory of response shift (Sprangers & Schwartz, 1999). This theory states that a change in a patient's perceived quality of life (QoL) (like the improvement in HRQoL that is often seen in chronically ill patients) is a result of an accommodation process. In this process, a "response shift" acts as a mediator and involves changing internal standards, values, and conceptualization of QoL. According to the theory, a response shift results from the interaction of several variables (e.g. changes in the health status, mechanisms such as social comparisons, and antecedents such as personality).

Fourth, several authors e.g. (Moss & Rosser, 2008; Rumsey & Harcourt, 2004; 2005, Chapter 4) point out that times of transition, especially those resulting in a change of social environment (e.g. changing schools, neighborhood or job), are particularly challenging for individuals with visible disfigurements. Because the scars of a corrected hypospadias are only visible in special situations such as sexual activities or in the locker-room after sports, childhood and adolescence may be particularly stressful for hypospadias patients. For example, in adolescence individuals may have their first sexual experiences and start their first intimate relationships. In adulthood, individuals are still confronted witch changing situations. However, by then many individuals are in a stable relationship or at least have had their first sexual experiences.

A fifth reason for the improving HRQoL of hypospadias patients over time may be their development of appropriate coping strategies. However, such a development might be difficult, as the patients' penile condition is not visible to others in most situations. One might expect that a less visible malformation is associated with a lower level of distress than everyday visible disfigurements. Yet, because a generally visible disfigurement may consistently provoke negative reactions from others, these individuals are somehow forced to develop ways of coping and to adapt to their appearance (Moss & Rosser, 2008; Rumsey & Harcourt, 2004; Thompson & Kent, 2001). In contrast, individuals with a less visible disfigurement may have feelings of a lack of control due to a greater variability of reactions

by others, and this may cause much more stress (Rumsey & Harcourt, 2004). In particular, individuals with concealable disfigurements (including conditions such as hypospadias that are only visible in certain situations) might avoid anxiety- or shame-triggering situations and thus have limited opportunities to develop positive ways of coping. Although concealment of a body part may temporarily reduce stress, such concealment might cause anxious thoughts that the hidden part will be revealed (Moss & Rosser, 2008; Rumsey & Harcourt, 2005, Chapter 5; Thompson & Kent, 2001). In line with this, studies on hypospadias have shown that many men with corrected hypospadias are afraid or ashamed of undressing in front of others (Rynja, de Jong, et al., 2011). To our knowledge, only one study (however only summarized in a congress abstract) has examined which type of coping is mainly used by individuals with corrected hypospadias (Rynja, De Kort, et al., 2011). It showed that men with corrected hypospadias more often avoided certain situations than a reference group of the Dutch population. This avoiding coping style may be problematic, as its application prevents the learning of new coping strategies and the disconfirmation of anticipatory anxious thoughts (Thompson & Kent, 2001). However, after individuals with corrected hypospadias have gained their first sexual experiences with someone else, it is possible that their unrealistic expectations of being ridiculed and rejected because of their penile appearance may be disconfirmed. This may contribute to the normalized HRQoL we have found in adults.

1.2.2. Determinants of HRQoL in men living with corrected hypospadias

Although our results indicated that adults with corrected hypospadias have a normal HRQoL, the quality of life reported varied between the individuals interviewed. Therefore, we were interested in which factors predict better and which worse physical and mental dimensions of HRQoL in men with corrected hypospadias. The following determinants were examined: age, living in an intimate relationship, severity of hypospadias, genital self-perception (PPS), orgasmic function (OF) and erectile function (EF).

Whereas none of the variables turned out to be a predictor of physical HRQoL (PCS), a worse genital self-perception (PPS), and a low orgasmic function (OF) were risk factors for impaired mental HRQoL (MCS).

Results concerning genital self-perception are congruent with the study by Schonbucher et al. (2008a), who found a significant positive correlation between mother-reported child HRQoL and penile self-perception in older children. Furthermore, our results are in line with those from studies on psychosocial adjustment, a topic that is related to mental dimensions of HRQoL. These studies (Mureau et al., 1997) found a better genital self-perception to be associated with better psychosocial functioning. However, correlations were small, and the significant ones were found for younger hypospadias patients (children and adolescents).

Finally, an association between genital self-perception and sexual satisfaction (which can be regarded as an aspect of quality of life) was found in men with a corrected hypospadias (Aulagne et al., 2010).

In our study, orgasmic function turned out to be the most significant risk factor for an impaired mental HRQoL. As problems with ejaculation (Rynja, de Jong, et al., 2011), a lower orgasmic function (Fraumann et al., 2014; Rynja et al., 2009), and a negative genital self-perception (Rynja, de Jong, et al., 2011) are mentioned more often by men with a corrected hypospadias than controls and ejaculation difficulties and a bad surgical outcome are more common in obsolete surgical techniques, it is possible that both variables are no longer risk factors for patients operated nowadays. However, we have to consider that genital self-perception does not always correlate with the objective cosmetic surgical outcome (Mureau et al., 1996). Therefore, even better surgical techniques probably do not consistently improve the genital self-perception of men with corrected hypospadias and therefore lead to improved HRQoL.

In contrast to our hypotheses, severity of hypospadias predicted neither mental nor physical HRQoL. However, this result is perfectly in line with the findings of Schonbucher et al. (2008a), who also showed that severity of hypospadias was not a risk factor for HRQoL in children and adolescents. In addition, Chertin et al. (2013) found that the type of hypospadias was not associated with the mental or physical HRQoL reported by their adult patients.

Interestingly, studies regarding different types of disfigurements also found that the severity of a disfigurement did not predict adjustment (Rumsey & Harcourt, 2004). Overall, it seems that psychological variables of hypospadias (such as genital self-perception) have a greater significance for the prediction of HRQoL than medical, hypospadias-related characteristics such as severity. Similarly, patients' well-being correlates with their subjective evaluation of the hypospadias and not with the objectively assessed surgical result (Aulagne et al., 2010). Also studies of children who have had an accident or have been chronically ill showed that social parameters are more likely to explain differences in HRQoL than medical parameters (Landolt & Sennhauser, 2007).

Contrary to our results, a study with children and adolescents (Schonbucher et al., 2008a), found higher age to be a predictor of a better HRQoL. It is possible that, in contrast to that age group, the sample of men in the present study (18-41 years) has a quality of life that remains relatively stable despite increasing age, particularly, because we have excluded participants with condition that affect the physical or mental health of an individual and cannot be

corrected or had not been corrected until this study was performed. It may be that all diseases were excluded which normally occur with age and affect the quality of life.

As discussed before, HRQoL seems to change between childhood and adulthood. We speculated that perhaps the HRQoL normalizes as soon as individuals with corrected hypospadias have become sexually active and have had their first experiences within an intimate relationship. Most of our participants were sexually experienced and thus possibly have had at least one intimate relationship. This might be the reason that the variable "being in an intimate relationship" did not turn out to be a predictor of their HRQoL.

Astonishingly, not even erectile function, when measured with the International Index of Erectile Function (IIEF) (Rosen et al., 1997), was a predictor of any dimension of HRQoL, although recent studies (Rynja, de Jong, et al., 2011) have shown that erectile dysfunction is more prevalent in hypospadias patients than controls. Furthermore, a study among men without a genital malformation also showed that erectile dysfunction was associated with impaired HRQoL (Sanchez-Cruz et al., 2003). However, since our sample of hypospadias patients had good erectile function, comparable to that of circumcised men, and no-one reported severe or complete erectile dysfunction, it is possible that this variable would be a predictor of HRQoL in another sample of individuals with hypospadias.

Besides our multivariate analysis, we also investigated whether medical variables were related to patients' HRQoL. In accordance with Schonbucher et al. (2008a) and Weber et al. (2009), we could not find any significant correlation between age at operation and HRQoL. This is interesting, because theories on psychological development (Schultz et al., 1983), the personal experience of many surgeons (Manzoni et al., 2004) and the American Academy of Pediatrics (1996) suggest that operations around the first birthday should have the smallest negative influence on HRQoL, although these notions lack supporting evidence.

2. Limitations of the current research project and considerations for future research

2.1. Limitations of study A and considerations for future research

This is the first cross-sectional analysis of genital perception of HASRGs by laypersons. The strengths of the current study are its reference to a theoretical background, the comprehensive study design, and the use of multivariate statistics. Regarding the study design, only standardized pictures of former hypospadias patients and a control group were used for the questionnaire. Finding hypospadias patients and circumcised men who accepted photo documentation of their penis for the questionnaire was a major challenge.

Nevertheless, some limitations have to be mentioned.

First, the small response rate of the volunteers for photographs might have biased the results. Possibly, mainly hypospadias patients who were satisfied with the surgical result of their correction participated, while those with a worse outcome probably did not attend. However, the urologists among the authors did not consider the results in photosets to be particularly favorable. On the contrary, they assume that patients treated with current techniques have better long-term outcomes than those in the study, who were operated up to 32 years ago.

Second, it is likely that the selected photosets do not represent the whole variation in outcomes of hypospadias repair. However, the selected photosets do represent a wide spectrum of different types of hypospadias.

Third, as in any survey on sexuality, selection bias is such that persons who are open-minded about that particular issue may be overrepresented (especially with the low -response rate of the participating laypersons) (e.g. Bogaert, 1996; Strassberg & Lowe, 1995). Besides, as laypersons with middle and upper socio-economic status (SES) and those in an intimate relationship were overrepresented, the results must be generalized carefully.

Fourth, we should consider that circumcision is not very common in Switzerland. As pictures of hypospadias patients were compared with those of circumcised men, the current results do not allow generalization to non-circumcised men. Therefore, future research may replicate our study and compare the genitals of men with corrected hypospadias with that of non-circumcised men without any genital malformation.

Fifth, our questionnaire contained more photos of shaved HASRGs than of shaved circumcised penises. This may have influenced laypersons' ratings, as shaving of pubic hair has been found to be an important genital aspect (Ramsey, Sweeney, Fraser, & Oades, 2009).

Sixth, we have to bear in mind that our data were collected cross-sectionally. Therefore, our results do not allow causal conclusions. For example, the positive effect of higher age in relation to laypersons' genital perception may be a result of the different generations and not of increasing age.

Seventh, the "mean score of laypersons' genital perception" was not validated.

Eighth, only photosets of men with corrected hypospadias were presented to women, and no photosets of men with non - corrected hypospadias. Therefore, the meatus might well be evaluated as more important by women when the questionnaire contained such photosets of non-corrected hypospadias.

As mentioned above at several points, research on genital perception in men with corrected hypospadias does not refer to any construct or psychological theory of genital perception. This is a major limitation of the literature, and to our knowledge our study was the first with a theoretical background regarding genital perception in men with corrected hypospadias. However, we cannot fully exclude the possibility that this doctoral thesis missed some relevant theories. Certainly, there are also other appropriate theories regarding the development of genital perception such as evolutionary and psychoanalytical approaches or such regarding the perception of disfigured people (Kent, 2000; Thompson, 2012). Besides, in the area of genital perception and reactions to penile appearance by others, the concept of stigmatization (Goffman, 1963) may be of significant importance. However, as a hypospadias is not visible in most situations, and as surgical correction achieves objectively good results, we believe that hypospadias patients are usually not stigmatized. In chapter A 2.3.2., we described the reasons for choosing Cash's CB model to explain the development of genital self-perception. Moreover, we cannot fully exclude the possibility that this thesis failed to consider a study concerning genital perception of men with corrected hypospadias. However, the most important findings are discussed.

For future research, we strongly recommend reference to a theoretical background. This will allow results and relevant conclusions to become comparable with future research.

Finally, although our results indicated that laypersons notice a difference between penile or more proximal types of HASRGs and circumcised genitals, it is not evident whether the

perceived difference bothers anyone. Therefore, we consider it essential that a future research also evaluates whether a perceived difference is bothersome.

Summing up, in our multiple regression analyses, the predictors of the dependent variables explained only a little of the variance. Dependent variables were "laypersons' genital perception of HASRGs" (in study 1), "perceiving HASRGs as normal-looking" (in study 2), and "perceiving circumcised genitals as normal-looking" (in study 2). There seem to be other, more important variables we did not include in our regression model. Thus, the importance of other predictors needs to be clarified in future studies.

It is possible that the factors which influence genital self-perception according to Cash's CB Model of body image (Cash, 2002, 2011) also have an influence on someone's genital perception of another person. Therefore, it can be used to generate future research questions regarding perceiver-related determinants of genital perception of HASRGs. As mentioned in chapter A 2.3.2.1, the CB model proposes four relevant influencing factors: cultural socialization, interpersonal experiences, physical characteristics and changes, and finally personality factors.

First, regarding the *cultural socialization* of the perceiver, it is possible that the genital perception of the perceiver is influenced by their concept of what a "perfect" penis looks like. As previously mentioned, this concept may differ between the different cohorts. For instance, as shaving of pubic hair was found to be an important genital aspect nowadays (Herbenick et al., 2013; Ramsey et al., 2009), younger laypersons may rate shaved genitals differently than older ones.

Second, regarding the *interpersonal experiences* of the perceiver, it is likely that values, attitudes and behaviors of the family and of peers regarding a malformation and genital appearance influence laypersons' rating of HASRGs. For example, when someone often hears evaluative comments of their friends regarding penile size, this issue may gain importance when evaluating penile appearance.

Third, regarding the *physical characteristics and changes* of the perceiver, men with a circumcised penis, and women who are in an intimate relationship with a circumcised partner, may have a different genital perception of HASRGs, due to the absence of the foreskin after hypospadias repair, compared to individuals that are not used to the appearance of a circumcised penis.

Another physical aspect that may influence women's rating of HASRGs is the menstrual cycle. As the menstrual phases of women can affect their evaluation of male faces (Johnston,

Hagel, Franklin, Fink, & Grammer, 2001), it is possible that they also affect the genital perception of HASRGs.

Fourth, regarding the *personality factors* of the perceiver, some factors that according to Cash contribute to the development of a positive or negative body image may also influence observers' genital perception of HASRGs. Suggested factors include self-esteem and perfectionism.

2.2. Limitations of study B and considerations for future research

Study B is the first study which assessed the HRQoL of adult patients with isolated hypospadias in comparison to a control group of circumcised men by means of a standardized and validated questionnaire.

However, there are some limitations. Although, they have already been described in section B3, the main limitations are mentioned again here.

First, the participation rate was rather low and the study had a limited statistical power. Therefore, it is possible that most of the study participants were individuals who had a positive approach to their corrected hypospadias and for whom the issue was not a taboo.

This could have biased our results towards an overly positive assessment of HRQoL.

Therefore, we recommend that a future study replicates the present pilot study with a larger sample size.

Second, it is possible that the generic HRQoL questionnaire which was used in our study might not be sensitive enough to measure concerns that are indicative of hypospadias. Therefore, the development of a disease-specific HRQoL measure for hypospadias patients should be considered (Schonbucher et al., 2008a).

Third, as already mentioned in the limitations of study A, we should remember that circumcision is not very common in Switzerland. As the HRQoL of our study sample was compared with that of circumcised men, the current results do not allow generalization to non-circumcised men. It is therefore important that a future study compares the HRQoL of men with corrected hypospadias with that of non-affected, non-circumcised men.

Fourth, since our data were collected cross-sectionally, our results do not allow causal conclusions. Therefore, future analyses of longitudinal data are needed.

Since case reports on men with non-operated hypospadias suggest that they adapt well to their penile condition, we further recommend that a future study not only replicates the present pilot study, which examined laypersons' genital perception and the HRQoL of men with corrected hypospadias compared to non-affected men. Notably, future research has to include men with non-operated hypospadias. One pilot study has already compared the sexuality of men with non-operated hypospadias with that of men without a genital malformation (Schlomer et al., 2014). However, this study did not investigate the HRQoL, and the main limitation is that participants identified themselves as having hypospadias. Therefore, a systematic investigation is needed into whether men with non-corrected hypospadias have a different HRQoL than men with corrected hypospadias or a control group of men without any genital malformation.

Summing up, in our multiple regression analyses a large proportion of the variance (especially that concerning the physical dimensions of HRQoL) could not be explained by the variables examined. Thus, the importance of other predictors needs to be clarified in future studies. Some suggestions for future studies for potential predictors and risk factors for HRQoL were made in chapter B 3. Besides, other variables that are determinants of HRQoL of children and adolescents with corrected hypospadias, the general population and disfigured people may also be of particular importance for the HRQoL of men with corrected hypospadias. Rumsey and Harcourt (2005, Chapter 5) divided factors into three groups which exacerbate or ameliorate distress in people with appearance concerns for example due to a disfigurement.

First, *physical and treatment-related factors*: In line with our findings, studies involving children and adolescents with corrected hypospadias, or individuals who have had an accident or have been chronically ill also found that these variables only have a limited influence on patients' HRQoL (Landolt & Sennhauser, 2007; Schonbucher et al., 2008a). However, the relationship between age at final operation and HRQoL in hypospadias patients should be re-examined due to its profound importance regarding the ideal age of hypospadias correction. This is particularly important, since our study and the study by Weber et al. (2009), which is the only research investigating the influence of age at surgery on HRQoL, had a purely explorative character with small sample sizes.

Second, *demographic and socio-cultural factors*: Since SES was associated with a subscale of the HRQoL (cognitive function) of children and adolescents with corrected hypospadias (Schonbucher et al., 2008a), the SES may be also a determinant of adult patients' HRQoL. Moreover, the influence of a patient's cultural context should be examined as suggested by Levine (1995).

Third, *psychological factors and processes*: These may involve, for instance, families' and peers' attitudes towards hypospadias such as the presence of potential worries in parents regarding a son's future sexuality or ridicule or rejections due to the penile condition by peers; families' or peers' social support of hypospadias patients; cognitive processes such as the interpretation of certain situations and adaptive or maladaptive coping styles; and finally personality traits such as self-esteem as suggested regarding psychosocial adjustment of disfigured individuals, an area related to mental dimensions of HRQoL (e.g. Rumsey & Harcourt, 2005, Chapter 5; Thompson & Kent, 2001).

In the search for variables that influence HRQoL, not only potential risk factors but also factors that provide resilience against the difficulties associated with hypospadias should be examined.

3. Clinical implications

The findings of the current PhD project on long-term psychological outcome of hypospadias repair allow some important conclusions for an improvement in hypospadias treatment.

Although our results indicated that men with corrected hypospadias have a normal HRQoL, our findings clearly showed that a negative genital self-perception and a lower orgasmic function were significant risk factors for an impaired mental HRQoL in hypospadias patients. In addition, men with corrected hypospadias were found to have a worse genital self-perception than non-affected men. Thus, hypospadias treatment should place more emphasis on the development of a better orgasmic function and of a positive genital self-perception. It can be assumed that an improvement in surgical techniques may result in better orgasmic function. However, it is not sure whether a reform of techniques would have the expected positive effect on patients' genital self-perception. Studies have shown that patients' genital self-perception does not always correlate with the objective cosmetic result (Mureau et al., 1996). Hence, there must be some other ways to improve patients' genital self-perception.

Cash's CB model (Cash, 2002, 2011), described in chapter A 2.3.2., not only offers a broad overview of the factors that might influence the development and maintenance of genital self-perception; it is also used to generate suggestions for improving patients' genital self-perception. The following clinical implications are based on the four factors of Cash's CB model that probably influence genital self-perception. Suggestions are made in relation to patients' social environment (e.g. parents and surgeons), patients' personality, cultural socialization, and surgical correction. The focus of these clinical implications is not only on promoting a more positive genital self-perception in hypospadias patients but also on improving hypospadias treatment generally.

Consideration of the social environment of hypospadias patients provides some suggestions for parents, surgeons and the medical personnel how to prevent the development of a negative genital self-perception and how to promote a positive one.

As mentioned in chapter A 2.3.2.2., several authors emphasize the profound influence of parents' feelings and attitudes towards the hypospadias of their child. They suggest that parents' emotions might be transmitted to their sons (Denson & Terry, 1988; Easson, 1966) and thus influence their genital self-perception. Actually, parents of children with hypospadias are often concerned about their son's sexuality (Metzler et al., 2014) and may feel guilty because of their son's malformation (Cobbett, 1974; Robertson & Walker, 1975). In addition,

an analysis of online hypospadias support groups (Springer, Reck, Huber, & Horcher, 2011) indicated that parents of hypospadias-affected children have a lot of unanswered questions concerning hypospadias. Therefore, a comprehensive counseling by the surgeon is of great importance to answer such questions and to eliminate or reduce any concerns which otherwise might be transmitted to the child. Besides information about etiology, genetics, fertility issues, and surgical techniques, such a counselling should also include new insights into psychological issues. Families should be particularly aware that their attitude towards the hypospadias and their information about malformations to the child may be as relevant as the surgical result. In addition, the communication of our present findings may reduce parents' concerns regarding a hypospadias-affected child's future. For instance, parents (and the child if already able to speak) should know that most types of HASRGs are not visibly different for laypersons and that the HRQoL of adults with corrected hypospadias seems to be normal. In addition, they should be informed that women tend to look at overall genital appearance and not at individual penile aspects such as the position and shape of the meatus or penile size.

Besides a comprehensive counseling of parents, we recommend a late follow-up in adolescence or beyond puberty, as has been suggested by several authors (e.g. Bracka, 1989; Ekmark et al., 2013; Mureau et al., 1996; Schonbucher et al., 2008b; Springer, 2014). This ensures that patients, who are usually operated at an age when they are not able to understand counseling, receive relevant information from a professional and have the opportunity to ask any question or address concerns regarding their genitals. In adolescence in particular, young people begin to compare themselves with others and worrying about what others think. Therefore, it is particularly important that patients are informed about our current results at that age. This information should include, for instance, the fact that women consider the penile appearance of HASRGs to be as normal-looking as that of non-affected genitals, and that women rather look at the overall genital appearance and not at individual penile aspects such as the position and shape of the meatus or penile size.

Various studies have indicated the importance of comprehensive information (e.g. Mondaini, Ponchietti, Bonafè, et al., 2002; Schonbucher et al., 2008b) and of a late follow-up (e.g. Aho et al., 1997; Bracka, 1989; Ekmark et al., 2013) for good adaptation to the penile condition.

Families may have different strategies for dealing with a family member's hypospadias. However, they should be aware that openness and willingness to discuss issues in relation to the penile condition may help the child to negotiate his feelings (Bellew, 2012) and to develop a positive genital self-perception (Schonbucher et al., 2008b). Moreover, literature concerning disfigured patients (Rumsey & Harcourt, 2005, Chapter 4) indicates that parents may develop an overprotective parenting style. Therefore, they should be encouraged to foster independence and autonomy of their child. However, a resource-oriented approach should

place the focus in counselling on parents' strengths rather than ameliorating presumed deficits in their parenting style.

As the genitals of hypospadias patients are not usually visible to others, the reactions and attitudes of both parents and surgeons and the medical personnel to patients' penile condition is of significant importance, as it is part of the limited feedback patients receive. Therefore, surgeons and medical personnel should try not only to discuss any concerns and possible further surgical options but also to reinforce patients in their masculinity by giving them positive feedback on their penile appearance. They should also be aware that reactions of healthcare professionals to a malformation have a profound impact on parents' attitude towards the malformation (Rumsey & Harcourt, 2005, Chapter 4).

According to Cash's CB model (Cash, 2002, 2011), certain personality traits (e.g. self-esteem and perfectionism) influence hypospadias patients' genital self-perception. Although to our knowledge no studies have examined the relation between personality traits and genital self-perception in hypospadias patients, some findings in individuals without genital malformations (Bramwell & Morland, 2009) have revealed an association between self-esteem and satisfaction with one's own genital appearance. Therefore, if urologists notice a low self-esteem due to the penile condition or a negative genital self-perception, and if they are in doubt regarding a patient's well-being, they may recommend psychological assessment and treatment, as already suggested by Schonbucher et al. (2008a). As a study has indicated that hypospadias patients often use poor coping strategies (Rynja, De Kort, et al., 2011), a social interaction skill training may have some beneficial effects. In addition, cognitive-behavioral approaches which, for example, modify dysfunctional thoughts may help hypospadias patients to improve their genital self-perception.

Both, social interaction skill training (Moss & Rosser, 2008; Rumsey & Harcourt, 2004; Thompson & Kent, 2001) and cognitive-behavioral approaches were found to be effective in the related area of disfigured patients (Rumsey & Harcourt, 2004; Thompson & Kent, 2001).

Unfortunately, although adults with a corrected hypospadias more often suffer from depression and anxiety than non-affected adults (e.g. Wang et al., 2010), no routinely offered support or intervention is available. As any psychological interventions are reactive rather than preventive, a routine assessment of patients' levels of psychological distress should be included in patients' follow-up care, as recommended by Rumsey and Harcourt (2004) in the related area of disfigured children. Such a measure should be short in length, easy to apply, and able to reliably identify patients who require specialist support (Rumsey & Harcourt, 2004). Thus, such a screening instrument needs to be developed in the future.

Cash's model (Cash, 2002, 2011) states that cultural norms influence our opinion of what is a good-looking or normal-looking penis. Several studies have indicated the correlation of cultural norms (e.g. transmitted by pornography) and genital self-perception (Mondaini, Ponchietti, Gontero, et al., 2002; Morrison et al., 2006). Some authors also suggest an association between cultural norms and aesthetic genital surgery (e.g. penile augmentation) (Mondaini, Ponchietti, Gontero, et al., 2002). Thus, such norms have a powerful impact on whether an individual with a corrected hypospadias feels that he has a normal or abnormal penile appearance and possibly whether parents decide to surgically correct a mild type of hypospadias or not.

Interventions should therefore not only be targeted at the hypospadias patients, but also at society in general by changing normative values. Such efforts can be undertaken at several levels. First, in accordance with authors in the related area of disfigured individuals (Rumsey & Harcourt, 2004), we recommend reconsidering the negative terminology used in relation to hypospadias. The terms "malformation" or "anomaly", which are commonly used to describe hypospadias (Baskin & Ebbers, 2006; Schonbucher et al., 2008a) could be replaced by neutral descriptions such as "variations" or "differences". A less negative terminology would certainly have a positive influence on patients' feelings about having a hypospadias.

Second, media regarding penile development and penile appearance (e.g. biological books, magazines in relation to sexual education or TV-documentary) should illustrate a broad range of different penises to represent the wide variation of "normal" penile appearance.

Third, for a change in normative values regarding penile appearance of individuals with hypospadias, more papers are needed such as that by Fichtner et al. (1995), who emphasize the wide variation of normal meatal location.

Such interventions may help individuals with corrected and non-corrected hypospadias to feel normal and socially accepted.

The present results also allow some considerations regarding surgical correction of hypospadias. To reduce complications after reconstructive surgery of severe forms of hypospadias which arise as a consequence from moving the meatus to the tip of the glans, in line with Fichtner et al. (1995) we recommend not to insist on moving the meatus as close to the tip as possible if it cannot easily be performed. With Mureau et al. (1996), we furthermore suggest that a reoperation of a retracted meatus is only indicated if requested by the patient himself.

Finally, as already mentioned a few times, it is assumed that early surgery optimizes patients' psychosexual development and heath-related quality of life (HRQoL) although there is no scientific evidence (American Academy of Pediatrics, 1996). Contrary to this assumption, the HRQoL of boys and adolescents with surgically corrected hypospadias was found to be significantly impaired compared to non-affected controls (Schonbucher et al., 2008a). Although in the present research project and in the study by Chertin et al. (2013) adults with corrected hypospadias showed a HRQoL in a normal range, many adults with corrected hypospadias still have a poor genital self-perception (Rynja, de Jong, et al., 2011). Furthermore, this emerged as a risk factor for impaired mental HRQoL. Moreover, in line with Weber et al. (2009), we found that age at operation did not correlate with patients' HRQoL. This arises the question whether a subgroup of men with distal hypospadias may also be corrected at an age when a child is able to decide whether he wants his hypospadias corrected or not. Interestingly, some case reports about non-operated hypospadias showed that the malformation was not recognized either by many of the patients (Dodds et al., 2008; Fichtner et al., 1995; Katib, 2013) or by their sexual partners (Fichtner et al., 1995), or even by professionals (Donaruma-Kwoh et al., 2010). In mild forms of hypospadias, functional deficits are rare (Fichtner et al., 1995), and mild types are usually operated to achieve a slit-like meatus at the tip of the glans. However, hypospadias repair can lead to complications, and the treatment is assumed to be psychologically traumatic by patients (Bracka, 1999; Morgan & Mezey, 1999). Our study found that women rated the position and shape of the meatus as the least important aspect of penile appearance. In line with other authors (De Win et al., 2012; Dodds et al., 2008; Fichtner et al., 1995; Schlomer et al., 2014) the question arises whether all individuals with distal types of hypospadias benefit from routine surgical correction in early childhood. Alternatively, routine surgical correction in early childhood could be limited to severe forms that impair function, for instance. In mild forms, a simple circumcision could be considered as an alternative to a surgical reconstruction.

4. General conclusions

In summary, the central findings of the current PhD project do not support hypospadias patients' fear of being ridiculed or rejected by others because of their penile appearance. Laypersons unacquainted with hypospadias perceived corrected distal forms of hypospadias (which represent the majority of hypospadias) as satisfactory and as normal as circumcised genitals. More proximal forms of hypospadias had a more negative laypersons' genital perception and were evaluated as less normal by women than circumcised genitals. However, the difference does not appear to be clinically relevant as the effect size was small.

Regarding women's concept of what a penis should look like, our findings indicated that women rather look at the overall penile appearance than at single penile aspects such as the position and shape of the meatus or penile size.

Our data also suggest that the HRQoL of adult hypospadias patients is comparable to that of a control group of circumcised men.

As a consequence for clinical management, we suggest that hypospadias patients and their parents be informed about these results. This information might reduce possible worries of parents. Moreover, it would help to prevent the development of shame due to the penile appearance and the emergence of an impaired genital perception in the patients. The genital perception of HASRGs by laypersons may vary due to several influences. Notably, we found that HASRGs seem to be rated more positively when laypersons know more about the "normal" variation of penile appearance (e.g. with increasing age and with a higher sexual interest). Therefore, efforts can be undertaken at several levels to inform the general population about the variation of penile appearance (e.g. by illustrating a broad range of genital appearances in books in relation to sexual education).

Although our study found normal HRQoL in men with corrected hypospadias, a negative genital self-perception was found to be a risk factor of impaired mental HRQoL. In contrast, most medical variables such as the age at operation or severity of hypospadias did not correlate with patients' HRQoL. Therefore, future hypospadias treatment should not only focus on improving surgical techniques but also on psychologically supporting hypospadias patients and their parents (e.g. for the development of a positive genital perception).

The present research showed that, although mild types of hypospadias are mainly operated to achieve a slit-like meatus at the tip of the glans, women considered the position and shape of the meatus to be the least important aspect of a penis. This may stimulate reflections regarding the relevance of correction of the meatus in minor forms of hypospadias in early childhood, although the women participating in our study were not exposed to photos of non-corrected hypospadias. To know whether every mild type of hypospadias has to be surgically corrected around the first birthday, it is important for further studies not only to assess the HRQoL of adults with corrected hypospadias but also to investigate men with non-operated hypospadias.

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- Weber, D.M., Landolt, M.A., Gobet, R. & Greeff, N.K. *The Penile Perception Score: An instrument for urologist's to evaluate results and for self assessment after hypospadias repair.* 22nd Annual meeting of European Society of Paediatric Urology (ESPU), Copenhagen, April 27-30.

Prizes:

1st prize: Oral presentation	Ruppen- Greeff, N.K., Landolt, M.A., Gobet, R. & Weber, D.M. (2014). Penile size in men with corrected hypospadias. 25th Annual meeting of European Society of Paediatric Urology (ESPU), Innsbruck, May 7-10.
Creativity Prize	Ruppen-Greeff, N.K., Landolt, M.A., Gobet, R. & Weber, D.M. (2013). *What's a good looking penis?* 3rd Retreat of the Children's Research Centre, Zürich, October 31.
3 rd prize: (PRIX Nachwuchs)	Ruppen- Greeff, N.K., Landolt, M.A., Gobet, R. & Weber, D.M. (2013): *What's a good looking penis?* Annual meeting of the Swiss Society of Pediatric Surgery, Zürich, September 19-20.
1st prize: Oral presentation	Greeff, N.K., Landolt, M.A., Gobet, R. & Weber, D.M. (2013). *What's a good looking penis?* 24th Annual meeting of ESPU, Genoa, April 24-27.